ANNALS OF SURGERY

VOL. 117 MARCH, 1943 NO. 3

CONTINUOUS CAUDAL ANALGESIA IN SURGERY*

JAMES L. SOUTHWORTH, M.D., WALDO B. EDWARDS, M.D.,

AND

ROBERT A. HINGSON, M.D.

STAPLETON, N. Y.

FROM THE UNITED STATES PUBLIC HEALTH SERVICE, MARINE HOSPITAL, STAPLETON, STATEN ISLAND, N. Y.

CONTINUOUS CAUDAL ANALGESIA, a method for prolonged caudal block, has been found useful in several types of surgical procedures. In some instances it has proved to be the anesthetic of choice. It is the purpose of this communication to discuss the application of the method, to note our recent experience with it, and to outline a technic for its administration.

There are several advantages which make continuous caudal analgesia worthy of consideration for use in general surgery. It is inherently a safe procedure and is comparatively easy to administer and control. The normal physiologic processes of the patient are little disturbed. It will find its field of greatest usefulness in operations about the lower extremities and the perineum, particularly in aged, debilitated, or recently shocked patients. It is also useful in inguinal and femoral hernioplasty. Further, it would seem that this method is adaptable for use in the treatment of casualties both in civilian and military practice where it would be desirable to have a safe, prolonged analgesia during the transportation and the physical and roentgenologic examinations of the injured. One anesthetist with several trained corpsmen or medical attendants could block a large number of patients at one time. It may be possible in some instances to block certain casualties on the field and transport them painlessly to a base for treatment, thus, perhaps reducing the instances of shock. On theoretic grounds it would seem that this method might be of value in the prevention of the delayed shock condition usually referred to as "crush" syndrome. Finally, limited experience in this hospital has shown promise that results similar to those obtained by Ochsner with lumbar sympathetic block in the treatment of thrombophlebitis and embolism of the lower extremities may be obtained with this procedure.

This method has been employed by us, and our associates, in more than

* Published with permission of the Surgeon General of the United States Public Health Service.

255 cases in general surgery, 150 of which were previously reported before the October, 1942 meeting of the American Society of Anesthetists. Table I lists the types and number of operations.

In three of these cases there were failures of the analgesia to reach the desired somatic level. In one instance, when given for inguinal hernioplasty, with a properly placed needle, the effect of the drug did not extend high enough to block the ilio-inguinal nerve in spite of shortened interval of injection and increased amounts of the drug. In the other two cases the failure was due to improper placing of the needle.

In inguinal hernioplasty the relaxation of the musculature is comparable to that of an expertly administered field block and often exceeds that of spinal. Traction on the cord and peritoneum in ligating the sac does not cause pain. An increased tendency to bleeding from the cut skin edges is noted. This tendency is not noted in the muscle layers should a vessel there be inadvertently cut.

In plastic operations about the vagina and perineum, it is an ideal analgesic. There is little interruption of the patient's usual routine of living. Meals are seldom missed either before or after the operation. The muscle relaxation is adequate. The presence of motor function in the extremities during and immediately after the operation tends to promote movement on the part of the patient, a condition thought to favor the prevention of thrombosis and embolism. The analgesia was satisfactory in three patients operated upon abdominally for lesions in the pelvis and for one undergoing cesarean section.

TABLE I

TYPES AND NUMBER OF OPERATIONS

Types of Operations	No. of Operations
1. Inguinal and femoral hernioplasty	43
2. Plastic operations on the perineum	21
3. Cesarean section	1
4. Hemorrhoidectomy; plastic operations about the anus; and abdominal operations on pelvic organs	34
5. Open and closed reductions of fractures of the lower extremities	24
6. Orthopedic procedures on the lower extremities	18
7. Amputations of lower extremities	7
8. Prostatic resection and other urologic procedures	41
9. Phlebectomy and ligation and injection of varicose veins	35
10. Appendicectomy	2
11. Treatment of thrombophlebitis	29

Those who prefer spinal anesthesia, or who undertake prolonged and meticulous operations for hemorrhoids, will find this method satisfactory. The relaxation of the anal sphincter is marked. Our experience indicates that the analgesia obtained from an expertly administered continuous caudal block is just as satisfactory as the seven-needle technic of sacral caudal block.

In orthopedic procedures on the lower extremity the method has been found of value in the recently shocked or elderly patient.

In urologic operations the advantages of complete lower analgesia without the dangers inherent in spinal anesthesia are present.

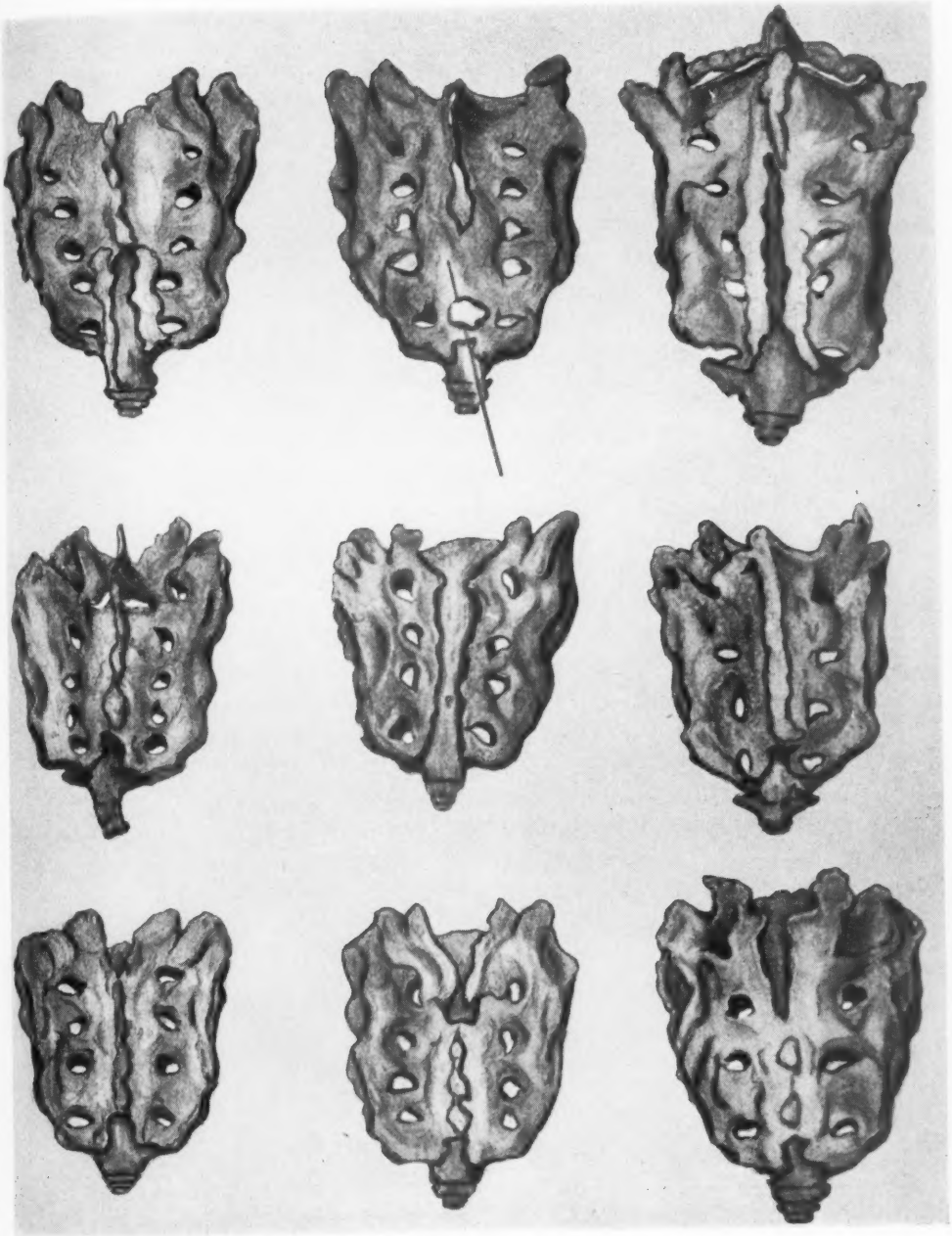


FIG. 1.—Common anomalies of the sacrum collected in the museum of anatomy from the Jefferson Medical College, Philadelphia, Pa. (courtesy Dr. George P. Pitkin: "Conduction Anesthesia," in preparation).

In phlebectomy an attempt is made to keep the dosage of the drug low so that the motor nerves are not affected. It has been found that the patient can aid the surgeon in maintaining position of the extremities during the operation.

With the present technic, the method is not recommended for appendicectomy, although in the two cases in this series it proved satisfactory.

In the treatment of thrombophlebitis promising results have been obtained. We have previously treated about 50 patients with the method of Ochsner in which lumbar sympathetic block is employed. The results were encouraging. The results in the patients treated with prolonged caudal block by the continuous method have been equally encouraging. It is our practice to maintain the block for four to six hours followed by a four-hour rest with the needle still in place. A second period of block of four hours duration is then administered. The following technic of administration is recommended for use in general surgery.

TECHNIC OF ADMINISTRATION

The patient is placed in the prone position and the sacral hiatus is palpated. The more common formations and anomalies of the sacrum, as shown in Figure 1, should be kept in mind. It is often of value to palpate the tip of the coccyx with the left forefinger and at the same time locate the sacral hiatus with the left thumb by palpation of the sacral cornua which lie on either side of the hiatus. The forefinger is moved to replace the thumb. The forefinger then serves as a guide to the location of the hiatus.

A continuous caudal set is assembled as shown in Plate 1. Since the use of the set shown, there have been no accidents involving broken needles in 200 consecutive injections.

The agent recommended is metycaine one and one-half per cent solution in physiologic saline. With this solution a skin wheal is raised just inferior to the hiatus. A special malleable No. 19-gauge needle is then inserted through the wheal and into the caudal hiatus, piercing, as it enters the canal, the sacrococcygeal ligament of Cathelin.

Careful aspiration should then be performed. If cerebrospinal fluid is withdrawn, it is recommended that the procedure be abandoned. This may occur in anomalous low-lying dural sacs. If bleeding through the needle is encountered, the needle should be withdrawn one to two centimeters and reinserted. When the bleeding has ceased the operator may proceed with the injection.

With the palm of the left hand firmly placed on the sacrum, ten cubic centimeters of the one and one-half per cent metycaine solution are injected. When the needle lies dorsal to the sacrum an "injection tumor" can usually be palpated. If the needle is correctly placed, the patient will complain of some unusual sensation in the lower extremities. This may be a transient ache, pain, or "shooting sensation" in the thighs or popliteal areas.

After the needle is properly placed, an average of 50 cc. of solution is

injected, according to the size of the patient. The analgesia begins in the areas supplied by the coccygeal nerves and progresses to those supplied by the hemorrhoidal, perineal, pudendal, ilio-inguinal and iliohypogastric nerves. Topographically, the analgesia usually begins about the coccyx and spreads in a circular manner up over the sacrum posteriorly and anteriorly over the perineum to the anterior abdominal wall. The toes begin to become insensitive to pain and the analgesia rises up the lower extremities. In the average patient analgesia will become complete from the umbilicus downward after 20 to 30 minutes.

A valuable sign that the needle is properly placed and the analgesia is progressing satisfactorily is the flushing of the toes and feet, which occurs within 5 to 15 minutes after the initial injection. Cold, moist feet become pink, warm, and dry.

Supplementary injections are given as required to maintain the desired analgesia. Twenty cubic centimeters every 30 to 40 minutes usually suffice. With proper precautions, the administering syringe may be placed near the field and supplementary injections may be administered by the surgeon or his assistant.

The work of the early investigators with single injection caudal analgesia showed that the height can be varied more or less in proportion to the speed of the injection and the amount of the fluid injected. We have found that when amounts larger than those recommended are used, and injected with more than the usual speed, analgesia can be obtained to the clavicles in many instances.

During the first 30 to 40 minutes after injection, the patient will not have complete loss of position sense although there will be absence of pain sensation to the umbilicus after 30 minutes. Increasing the amount of drug and decreasing the time interval between injections will cause a seepage of the agent anteriorly until the motor fibers are blocked. Patients subjected to the action of the drug, by this method, longer than one and one-half hours will also frequently exhibit this latter phenomenon.

The time consumed in the administration and the prolonged period during which the analgesia is incomplete constitute its most serious disadvantages. Thirty to 40 minutes are required to assemble the set, making the injection, and allow the analgesia to become complete. However, a skilled anesthetist, experienced in the method, can often have a patient ready for operation involving the perineum, vagina, or rectum in ten minutes.

As has been described in previous papers,[1, 2, 3, 4] the complications arising from the use of the method have not been of great consequence in our hands. An occasional patient may experience mild syncope following the initial injection. It is thought that this may occur from one of two causes: (1) Either a small amount of the drug may have been injected into a vein, of which there are many in the sacral canal; or (2) it may be caused by sudden pressure on the dura from the injection of a comparatively large amount of solution. In our experience, the phenomenon has been transient (10 to 20 seconds

duration) and no sequelae have attended its occurrence. One moderately severe infection occurred in our entire personal experience. There was a low grade cellulitis about the sacral hiatus but there was no evidence of nerve injury. No other patient showed the slightest infection.

Failures of the method to produce the desired analgesia are due, in most instances, to the needle's being out of the sacral canal. It is sometimes difficult for one not specially trained and experienced in block anesthesia to insert a malleable needle directly in the midline and into the sacral canal. There are several common sources of error: The needle may lie superficial to the sacrum; it may lie in the periosteum of the roof or floor of the canal; it may lie within a blood vessel or within the subarachnoid space; or the needle may bend and come to lie lateral to the sacrococcygeal junction.

If a vein has been pierced at the beginning of the injection, a small hematoma may form about certain nerve trunks, giving patchy areas of sensation. Other than the failure of the analgesia in these areas, this has not been attended with sequelae in our series.

CONCLUSIONS

A. Continuous caudal analgesia has been found to be a valuable procedure in the following types of cases:

1. Plastic operations about the rectum and perineum in the fields of proctology, urology, and gynecology.
2. Operations below the umbilicus in the aged and debilitated, in which other forms of anesthesia are contraindicated.
3. Surgical and orthopedic repair of traumatism of the lower extremities.
4. Thrombophlebitis of the lower extremities.
5. Femoral and inguinal hernioplasty.

B. Experience indicates that the method is one of easy administration, accurate control, and one that can be prolonged as long as necessary, with little fear of complications.

REFERENCES

[1] Hingson, Robert A., and Southworth, J. L.: Continuous Caudal Anesthesia. Am. J. Surg., **58,** 92–96, 1942.

[2] Edwards, W. B., and Hingson, R. A.: Continuous Caudal Anesthesia in Obstetrics. Am. J. Surg., **57,** 459–464, 1942.

[3] Hingson, R. A., and Edwards, W. B.: Continuous Caudal Anesthesia During Labor and Delivery. Anesthesia and Analgesia, **21,** 301–311, 1942.

[4] Hingson, R. A., and Edwards, W. B.: Continuous Caudal Analgesia in Obstetrics. J. A. M. A., **121,** No. 4, 225–228, January 23, 1943.

EARLY MORTALITY OF BURNS AS INFLUENCED BY RAPID TANNING AND BY TRANSFUSIONS

ROBERT ELMAN, M.D.
ST. LOUIS, MO.

FROM THE DEPARTMENT OF SURGERY, WASHINGTON UNIVERSITY MEDICAL SCHOOL, BARNES HOSPITAL AND ST. LOUIS CHILDREN'S AND CITY HOSPITALS, ST. LOUIS, MO.

THE PURPOSE of the present communication is to draw a few inferences from a statistical study of the early mortality on over 400 cases of severe burns, with special reference to the type of local therapy and to the use of blood and plasma transfusions. The results of this limited study apply only to the important problem of reducing the early mortality, a feature of the treatment of burns which often has been neglected in favor of factors which deal with the subsequent treatment of these injuries. No attempt will be made to review the extensive literature on burns in view of the excellent summary reported by Harkins.[1]

SELECTION OF CASES

In order to study only the more serious burns, all admissions of minor burns were excluded, *i.e.*, patients who were discharged to the Outpatient Department within 24 hours. Moreover, only patients whose end-result was known were included, *i.e.*, patients discharged to other hospitals were omitted from consideration. During the seven years from 1935 to 1941, inclusive, 369 cases treated at the St. Louis City Hospital were analyzed. In addition, 59 cases treated at the St. Louis Children's and Barnes Hospitals were reviewed briefly. The cases were divided first into two groups, depending upon the method of local treatment, *i.e.*, those in which tannic acid was used, and those in which no tannic acid was used. The latter group included a variety of local treatments such as the application of saline packs and soaks, vaselined gauze, various ointments such as pyrobalm, *etc.* Only a few cases were treated with triple dye, and these were included under the nontannic procedure. Local and systemic sulfonamide therapy was employed in many cases treated during 1941, but no special study was made of this fact.

The method of tanning employed should be emphasized; rapid coagulation was produced in each instance following cleansing of the burned area. This rapid coagulation was achieved by the alternate application of ten per cent silver nitrate and ten per cent tannic acid. In no instance was tannic acid jelly or the slower method of tanning with tannic acid alone used.

CLINICAL FINDINGS

A study of the cases analyzed was concerned largely with the influence on early mortality of (1) local therapy; (2) plasma and blood transfusions; and (3) priority in which local and systemic therapy was applied.

Influence of Tanning.—In the City Hospital an equal number of cases were treated by tanning in contrast to those treated without tanning; although the mortality was lower in the latter group, the difference is too slight to be of significance (Table I).

TABLE I

	Total Cases	Fatal Cases	Mortality
Rapid tanning	180	37	20%
Nontannic therapy	197	32	17%
Total cases	368	69	19%

A question may be asked in regard to the manner in which these cases were selected for local therapy. While it was difficult to determine always on what basis this selection was made, one fact tended to make the division rather accurate, *i.e.*, patients were admitted alternately on one of two different Services, and for various periods of time the policy of tanning was employed in one unit, whereas it was not used in another unit.

However, if one breaks down the 368 cases into yearly totals, the number of cases treated with tannic and nontannic therapy were equally divided during the years 1939 and 1940, and during these two years there was a striking difference in the mortality between these two methods of therapy (Table II).

TABLE II

	Total Cases	Fatal Cases	Mortality
1939—Tannic	31	4	13%
Nontannic	38	10	26%
1940—Tannic	25	2	8%
Nontannic	30	5	17%

Of interest, also, is the fact that two fatal cases in the tannic group in 1940 occurred at four and six hours, and received no plasma. This undoubtedly was a mistake in the priority of treatment, the local having preceded systemic therapy—a serious error which is discussed below.

The cases treated at Barnes and St. Louis Children's Hospitals were also analyzed in regard to a possible correlation between mortality and the use of tannic acid *versus* the nontannic acid therapy. Here the difference was significant and was similar to the experience at the City Hospital just noted (Table III).

TABLE III

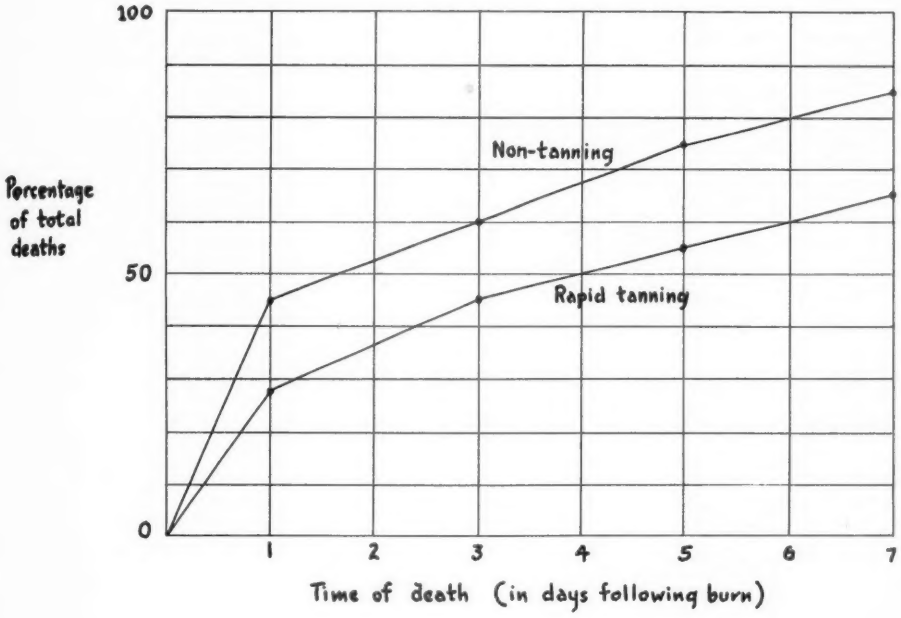
	Total Cases	Fatal Cases	Mortality
Tannic	15	1	7%
Nontannic	44	6	13%
Totals	59	7	11%

Of interest, too, was the fact that only one patient of the seven fatal cases received any blood or plasma.

The *time of death* in the St. Louis City Hospital cases was then analyzed, the fatal cases being again divided into two groups, one treated with and the other without tannic acid. The results were significant. It was apparent immediately that a smaller part of the patients treated with tannic acid died within the first few days than those treated without tannic acid. The difference has been plotted as a graph in Chart 1, and is really self-

explanatory. An obvious explanation of this difference is the effect of rapid coagulation in minimizing plasma loss. This inference is strengthened by the fact that, in more recent years, when large volumes of plasma were available, early deaths have diminished remarkably whether tannic acid was used or not. A second possible explanation is that the toxic absorption in cases burned, so as to produce wet, necrotic skin, was minimized by coagulation; this factor was the original basis upon which Davidson used tannic acid and, though difficult to prove, has, likewise, never been disproved.

CHART I



Influence of Plasma and Blood Transfusions.—That the use of adequate amounts of plasma has reduced mortality in severe burns was apparent from simple inspection of the mortality figures from year to year. The mortality at the St. Louis City Hospital was 25 per cent in the year 1936, and has dropped steadily to a level of 11 per cent in the year 1941. Mortality figures are, of course, notoriously inaccurate because of the great influence of the severity of the burn. Nevertheless, such a difference in mortality in six years during which the only consistent change in therapy was the increased use of plasma seems a definite indication of this influence. This change, involving the increased use of plasma, concerns three details which may be mentioned at this point: (a) The gradual displacement of the use of whole blood by plasma. (b) The increased amount used in severe cases, *i.e.*, as much as 5,000 cc. (c) The promptness with which plasma was used, made possible by the establishment of a plasma bank.

The influence of transfusions was even more strikingly evident when the fatalities at the St. Louis City Hospital were divided into two groups, *i.e.*, those treated with and those treated without plasma or blood. This analysis is listed in Table IV.

TABLE IV

Patients given no blood or plasma	25	
Death occurred within 24 hours	15	(60%)
Death occurred between 1 and 7 days	3	
Death occurred between 11 and 30 days	7	
Patients given varying amounts of blood or plasma	44	
Death occurred within 24 hours	6	(13%)
(of these, 3 received but 500 c.c. of whole blood, 1 received 1000 c.c. whole blood, and 2 each received 2000 c.c. of plasma)		
Death occurred between 1 and 7 days	21	
Death occurred between 8 and 90 days	17	

Note that, of the 25 fatal cases not receiving blood or plasma, 15, or 60 per cent, died within 24 hours, whereas only 6, or 13 per cent, of the 50 receiving this treatment died within 24 hours. Moreover, of the six cases in the latter group, four should really be listed as not receiving this form of therapy, inasmuch as the amount given was totally inadequate. If this transfer is made, it will be seen that only four per cent of the fatal cases given blood or plasma died within 24 hours, in contrast to 65 per cent in those receiving no blood or plasma, or inadequate amounts thereof.

The Influence of Priority of Treatment.—In analyzing the fatal cases in the present group, seven patients were found to have died within seven hours after admission. Study of these cases shows that the local treatment, often carried out under general anesthesia, was performed first and without adequate attention to systemic therapy, *i.e.*, the injection of plasma or blood. The inference, therefore, seems justified that certainly in a few of these instances, a fatality may have been avoided or at least postponed, if the treatment of shock were carried out immediately, and the local therapy delayed until the general condition had improved.

COMMENT.—The data analyzed in this study adds further emphasis to the importance of minimizing or correcting plasma loss *early* and *adequately*, if one is to lower early mortality due to shock. While a prompt and adequate plasma transfusion alone is quite satisfactory, rapid coagulation undoubtedly reduces the amount of plasma which is needed so that if the plasma transfusion is inadequate, this factor may tip the balance in favor of recovery. However, this is not the only beneficial effect which rapid coagulation probably has in reducing early mortality.

No one has yet disproved the original contention of Davidson that tannic acid, by coagulating dead tissue from which toxic products may be absorbed, minimizes or prevents the deleterious effects thereof. This theory depends, of course, upon the actual existence of *moist, necrotic* skin. The present writer has repeatedly observed such areas of skin in severe burns and believes that toxic absorption may occur whenever such a lesion is present just as easily as it can from nonviable intestine, from a wet gangrene of the leg, or

from any other moist, dead tissue in which autolysis occurs with or without the presence of infection.

The fact that absorption of tannic acid has been shown to produce evidence of toxic hepatitis would seem to offer a contradiction to the mechanism just described. This discrepancy, however, is more apparent than real. In the first place, it is hard to see how, during *rapid coagulation,* significant absorption could occur. During *slow coagulation* which follows the use of tannic acid jelly, or of solutions of tannic acid alone, such absorption might easily occur, as pointed out by McClure and Lam.[2] Significant is the fact that in four cases of toxic hepatitis reported by Wells, Humphrey and Coll[3] the slow method of tanning was used. On the other hand, Rhoads, Lee and Wolff[4] have described two cases (Cases 11 and 14) in which death occurred on the fifth day, the autopsy showing toxic hepatitis, but in which tannic acid was not used. The problem of toxemia in burns is far from clear, and obviously requires further study. Until definitely disproved, however, the original hypothesis of Davidson should be retained as a possible explanation of some of the early deaths in burns which still occur in spite of adequate control of shock with early and sufficient plasma transfusions.

SUMMARY

A statistical analysis was made of 78 fatal cases among 427 severe burns, with special reference to the probable influence of therapy on the early mortality. It seemed clear that the following three factors alone or in combination contributed to early mortality: (1) The use of noncoagulation methods of therapy. (2) Failure to use plasma transfusions at all, or in inadequate amounts. (3) Failure to precede local by systemic therapy.

The inference seems justified that the reduction of early mortality depends upon giving rigid priority to methods which minimize or correct plasma loss and which thus combat, early and rapidly, the systemic manifestations due to loss of plasma. The problem of toxemia as a cause of early deaths is discussed briefly.

REFERENCES

[1] Harkins, H. N.: The Treatment of Burns. C. C. Thomas, 1942.

[2] McClure, R. D., and Lam, C. R.: Military Surgical Manuals—V. Burns, Shock Wound Healing and Vascular Injuries. W. B. Saunders Co., 1943, page 30.

[3] Wells, D. B., Humphrey, H. D., and Coll, J. J.: New Eng. Jour. of Med., **26,** 629–635, April 16, 1942.

[4] Rhoads, J. E., Wolff, W. A., and Lee, W. E.: Annals of Surgery, **113,** 955–960, June, 1941.

GOITER INCISIONS

FRANK H. LAHEY, M.D.

BOSTON, MASS.

FROM THE DEPARTMENT OF SURGERY, THE LAHEY CLINIC, BOSTON, MASS.

A GOITER INCISION involves many more features than are at first apparent. The appearance of the operative scar, very properly and importantly, occupies the mind not only of nearly every patient, certainly every female patient, but also the mind of nearly every surgeon. However, this is by no means the only feature of importance in connection with a goiter incision. Since a goiter, particularly when associated with hyperthyroidism and intrathoracic goiter, possesses a high degree of possible fatality, it is most important that the incision provide complete and adequate anatomic exposure so that the surgeon can perform the operation without being hampered unnecessarily by technical complications, and with control of the thyroid's vascular supply. The method of making the incision and elevating the

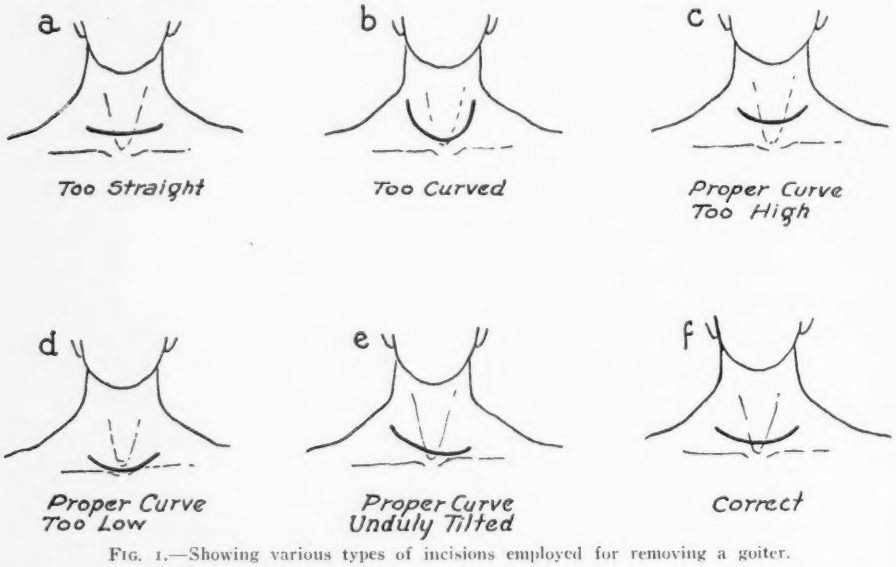


FIG. 1.—Showing various types of incisions employed for removing a goiter.

skin flap is also of great importance in what is often a serious procedure, since it influences the amount of bleeding and, therefore, also the length of time expended in controlling the bleeding and performing the operation.

Figure 1 a shows a goiter incision which is too straight and is unsightly because it cannot be concealed well by a necklace. A curved incision provides adequate exposure without particularly adding to the transverse length of the scar and so improves exposure.

Figure 1 b shows a goiter incision which is too much of the U- or horseshoe-type. The same amount of exposure in the part of the wound high

up beneath the skin flap can be accomplished with the symmetrical, slightly curved incision just above the junction of the neck with the chest without the unsightliness of this U-type of incision.

Figure 1 c shows a properly curved incision, but because it is placed too high it cannot be concealed satisfactorily by a necklace and is unsightly.

Figure 1 d shows a properly placed incision, but it is so low that it, likewise, cannot be concealed satisfactorily by a necklace.

FIG. 2.

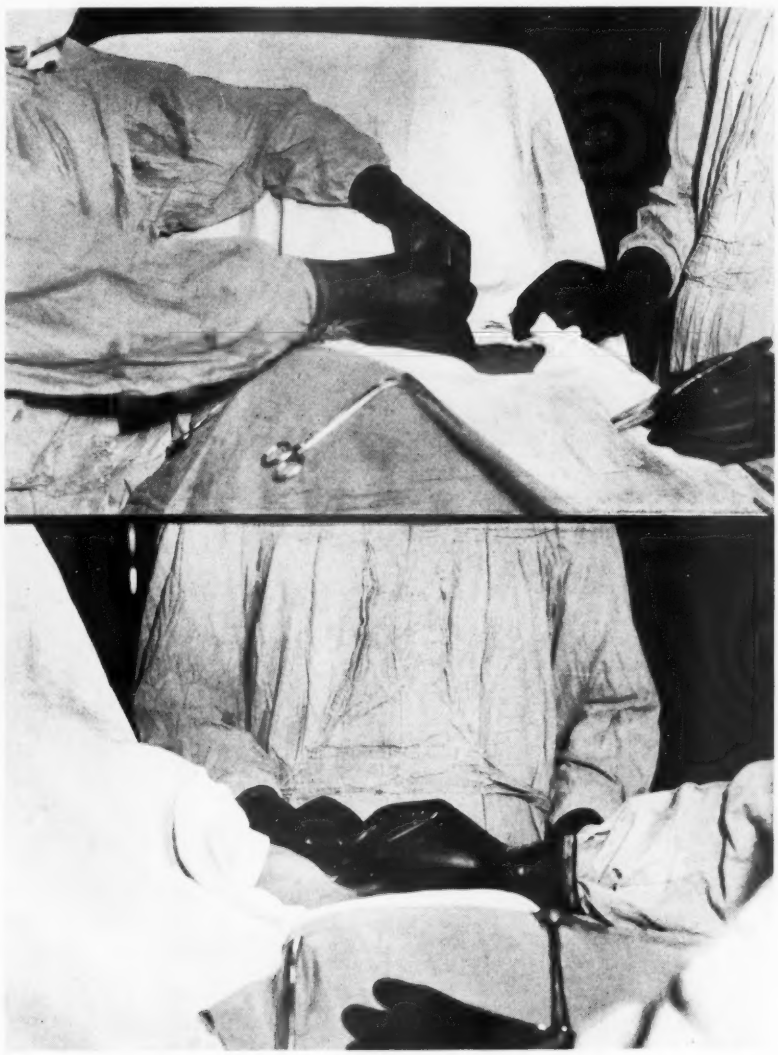


FIG. 3.

FIG. 2.—The surgeon stands with his back to the patient's head in the proper position to make a well balanced, well curved goiter incision without having to swing his body.

FIG. 3.—When the surgeon faces the patient's head, it is difficult to keep the incision in balance, and the right side of the incision may be lower than the left. This is the position so often taken by those inexperienced with thyroid surgery, in order to visualize the neck.

Figure 1 e shows a properly placed and curved incision, but because it is improperly tilted and out of balance it is unsightly.

Figure 1 f shows a properly placed and curved incision of adequate length for exposure of all anatomic structures. Note that the curve corresponds with the level at which a necklace will hang, that is, just above the point where the neck joins the chest, which is the level at which the incision is least noticeable.

I have often been impressed when seeing a patient upon whom we have performed a subtotal thyroidectomy, the patient having had auricular fibrillation and cardiac decompensation and having been almost bedridden, that the first comment frequently is: "All my friends are so impressed with the beauty

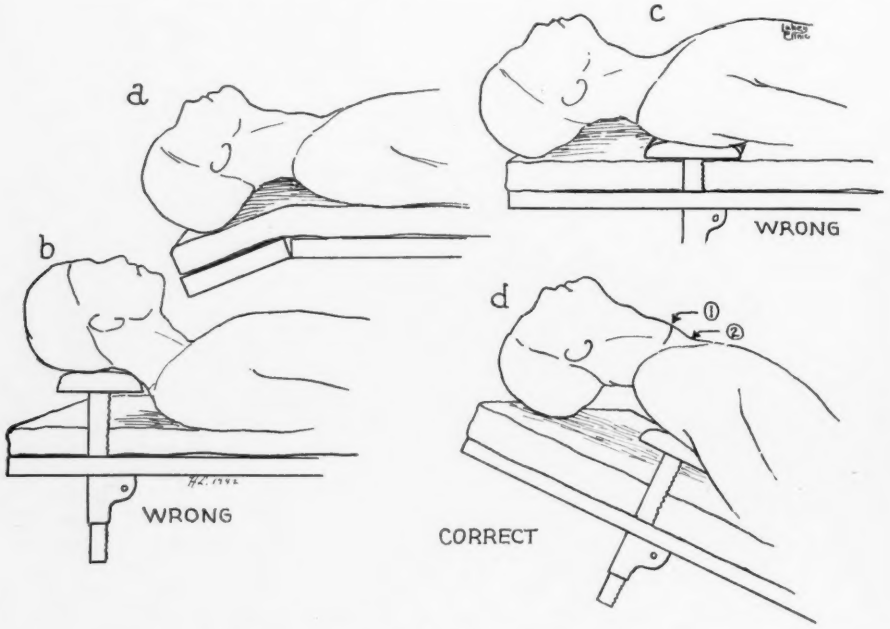


FIG. 4.—(a) The forward projection of the neck is inadequate when the head is dropped over the end of the table. (b) The goiter bar improperly placed beneath the neck, causing flexion of the neck on the wound. (c) The goiter bar properly placed well down beneath the shoulder blades. (d) The goiter bar properly placed beneath the shoulder blades. Forward projection of the thyroid can be obtained by elevating the chest. Arrow 1 shows where the incision must be made if it eventually is to be located at the point indicated by arrow 2, to which it will slip when the goiter bar is let down.

of this scar." In the meantime, normal rhythm has been restored, compensation retained, and all activities resumed, but elicitation of information concerning the physical improvement requires determined interrogation. This is not a critical observation, but indicates the necessity of satisfying the patient, and, after all, our obligation is to obtain the best possible scar in addition to the best possible exposure.

In the removal of 22,000 goiters we have learned many things about incisions. Not infrequently an assistant who comes to us inexperienced with goiter incisions is asked where he would make an incision. He often measures with the thickness of his fingers either one or two fingersbreadth

above the notch of the sternum and arbitrarily settles that as the point of incision. For a number of years we have taught assistants that a well placed goiter incision should not depend on such an arbitrary rule. There are long and short necks, which vary between the tip of the chin and the hyoid, between the notch of the thyroid cartilage and the notch in the sternum. The only way a goiter incision can be well placed is by a free-hand marking of the neck and by determining with the eye, coordinated with the hand, the most artistic location and the proper curve of the scar.

A properly placed and gracefully curved goiter incision must be made with the knife poised rather loosely in the fingers, with the arm relaxed, and with a confident sweeping curve. It cannot be made slowly and cautiously since this involves tension, irregularities in the sweep of the curve, and a tendency toward distortion in balance.

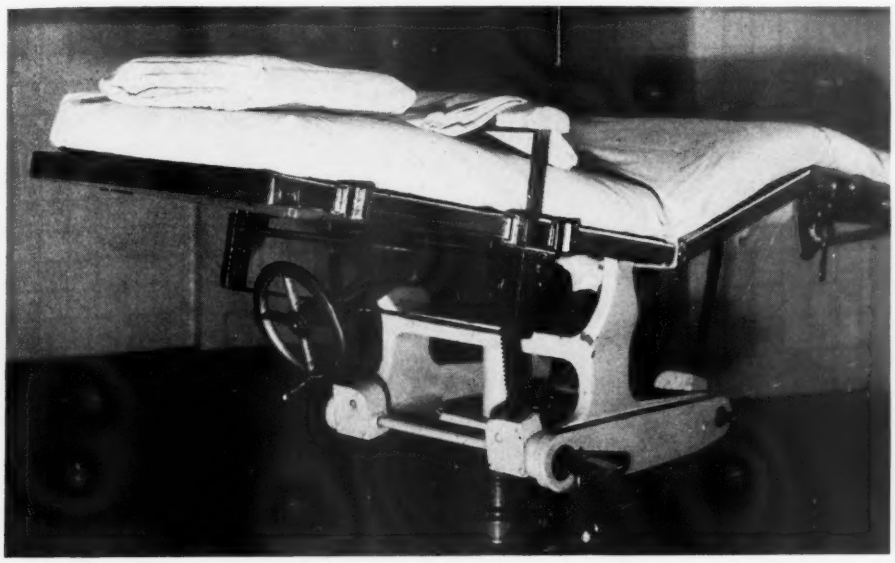


FIG. 5.—Goiter bar. Note that this goiter bar, designed at the Clinic, can be pushed away from or toward the head of the table, and is elevated by the wheel under the table.

There is a distinct advantage, in my experience, in standing with one's back to the patient's head in order to have a free arm and body turn, by means of which a curved incision can be completed as would be the case in free-hand drawing (Fig. 2). When the surgeon faces the patient's head and the knife is carried across the neck (Fig. 3), there is a tendency for the right side of the incision to be lower than the left. The two sides can be kept in balance only by compensatory rotation of the body or by twisting the wrist. Undoubtedly many surgeons can make a good incision while facing the patient's head, but it is easiest for me to keep the curve of the incision in proper balance if I stand with my back to the patient's head. Although this may seem like a small point, it does much to facilitate

a good incision not only for those surgeons who lack wide experience in thyroid surgery but also for those who have had extensive experience.

A forward projecting position of the neck in goiter operations is of the utmost importance not only in making a good incision but, also, in dealing technically with partial or complete removal of the thyroid gland. Attempts to obtain good exposure by permitting the head to drop over the end of the operating table or on a table, the upper end of which drops (Fig. 4 a), do not compare favorably with the results obtained with elevators properly placed beneath the shoulder blades (Fig. 4 c). Goiter bars, which permit

FIG. 6.

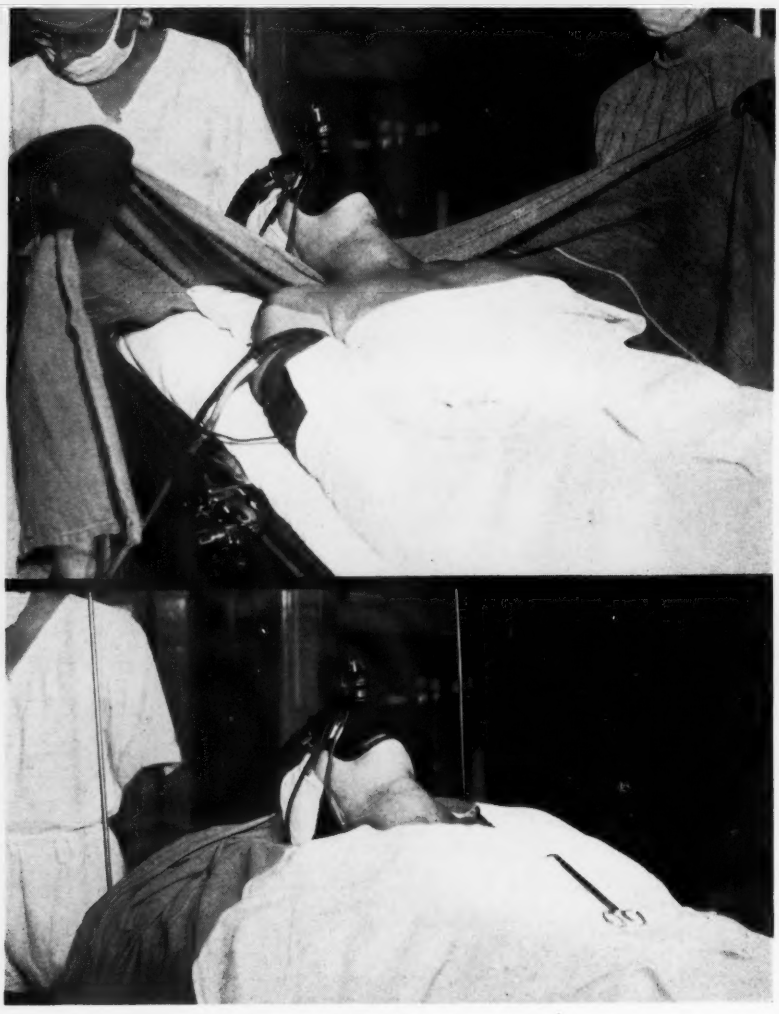


FIG. 7.

FIG. 6.—Draping for thyroid operation. The rubber tube connecting the anesthesia apparatus is removed, the head raised by the anesthetist, and a sterile sheet draped under the neck and head and carried down over the chest.

FIG. 7.—In the second step of draping, two towels forming a V are placed on the patient's chest and over the shoulders.

elevation of the neck, have existed for a good many years; however, they are often misused. The goiter bar should not be placed beneath the neck (Fig. 4 b), since in this position it forces the head forward, the chin downward, and hinders rather than aids the exposure of the thyroid gland. If it is properly placed well beneath the shoulder blades (Fig. 4 c and d), it throws the chest up and so widens the distance between the chin and the sternal notch that it throws the neck forward, puts the skin on the stretch

FIG. 8.

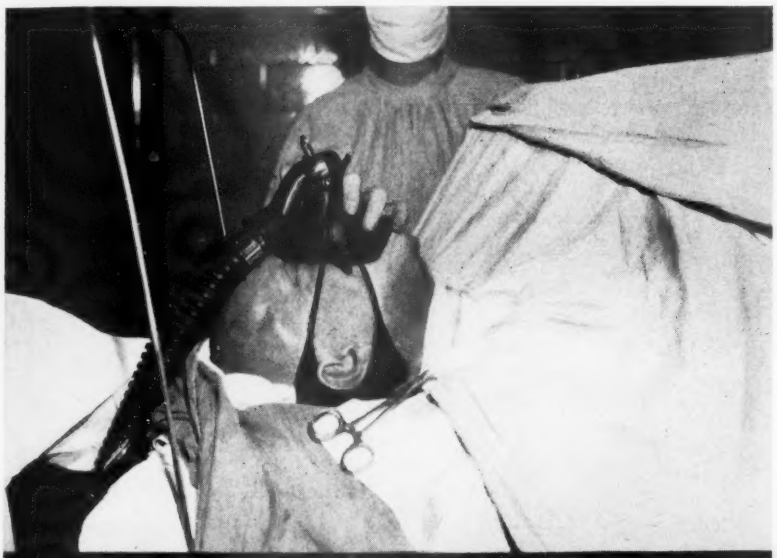


FIG. 9.

FIG. 8.—In the third step of draping, a sheet is clamped across the patient's neck, the towel clip catching this sheet, the edge of the two towels which form a V, and the edge of the sheet which was placed beneath the head and neck. When this sheet is turned upward (Fig. 9), the operative field is exposed.

FIG. 9.—The sheet is draped over the anesthetist's frame. A sheet with a U cut-out is now placed over the patient's body and chest. With the U clamped to the sheet and a towel to the sheet over the anesthesia frame, the operative field is now completely draped. With strips of gauze tucked down in the angles beside the neck, an adequate amount of neck and chest is exposed and the anesthetist completely excluded.

and forces the thyroid into the wound when the prethyroid muscles are severed. I mention this method of exposure further to warn that as the neck is arched forward, one must be careful to make the incision a little higher than one wants the scar to be because when the goiter bar is let down, the incision will descend toward the sternal notch from one-fourth to one-half inch (Fig. 4 d, 1 and 2).

Another important factor in a goiter incision is the exposure of a sufficient area of skin over the chest and up over the neck as the wound is draped, permitting the establishment of good visual relations as to the proper level for the incision. I have frequently seen a goiter incision made through a very small aperture in the towels, giving little opportunity to relate the extent and location of the incision to the remainder of the

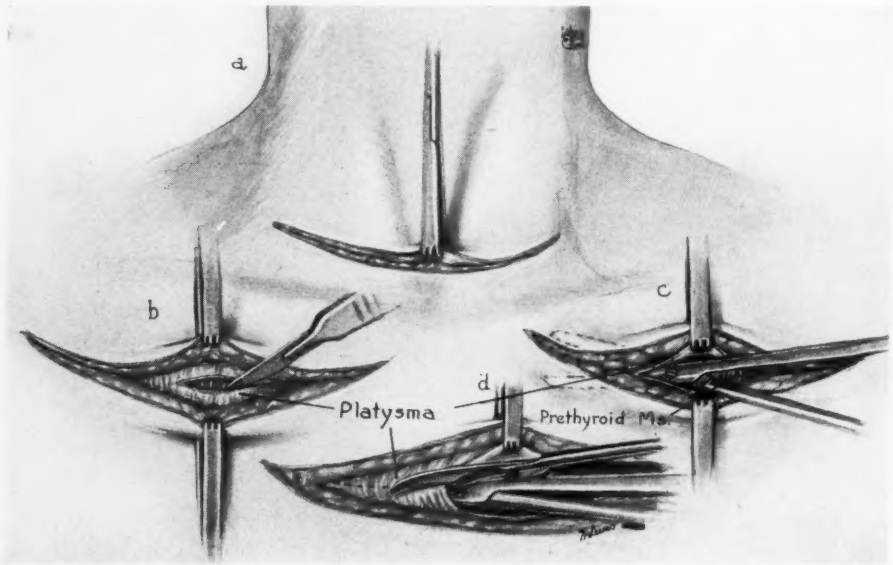


FIG. 10.—(a) Method of marking out the incision partly through the skin and not penetrating the entire thickness of the skin. (b) The incision is deepened until the platysma can be demonstrated and incised in the midline. (c) Metzenbaum scissors are inserted beneath the platysma to separate it from the prethyroid muscles. (d) The platysma separated from the underlying structures is now incised with scissors. This maintains intact the layer of adipose tissue between skin and platysma muscle.

neck and chest. In Figures 6, 7, 8 and 9 is shown the method utilized in this Clinic of draping thyroid patients. A good exposure of the neck is obtained, and the anesthetist is excluded completely from the operative field.

For many years we have taught that the first part of a goiter incision should extend only through the superficial one-half or two-thirds of the skin (Fig. 10 a); it can then be deepened in its central point (Fig. 10 b), through the skin, subcutaneous fat and platysma. As the platysma fibers are cut, platysma, subcutaneous fat and skin can be picked up above and below with double hooks. A pair of Metzenbaum scissors can then be introduced (Fig. 10 c) and opened beneath the platysma and in front of the prethyroid muscles so that the platysma is separated completely beneath both sides of the marked

out skin incision. The central portion of the incision which already has been deepened can then be elevated, and the marked out skin with the subcutaneous fat and the elevated platysma can be cut with scissors in one layer on each side (Fig. 10 d). This retains the attachment of the platysma to the subcutaneous fat, and double hooks are so applied between the skin and platysma that the platysma envelope enclosing subcutaneous fat is kept intact (Fig. 10 c and d). This layer just beneath the platysma demonstrates the proper avascular line of cleavage between the platysma

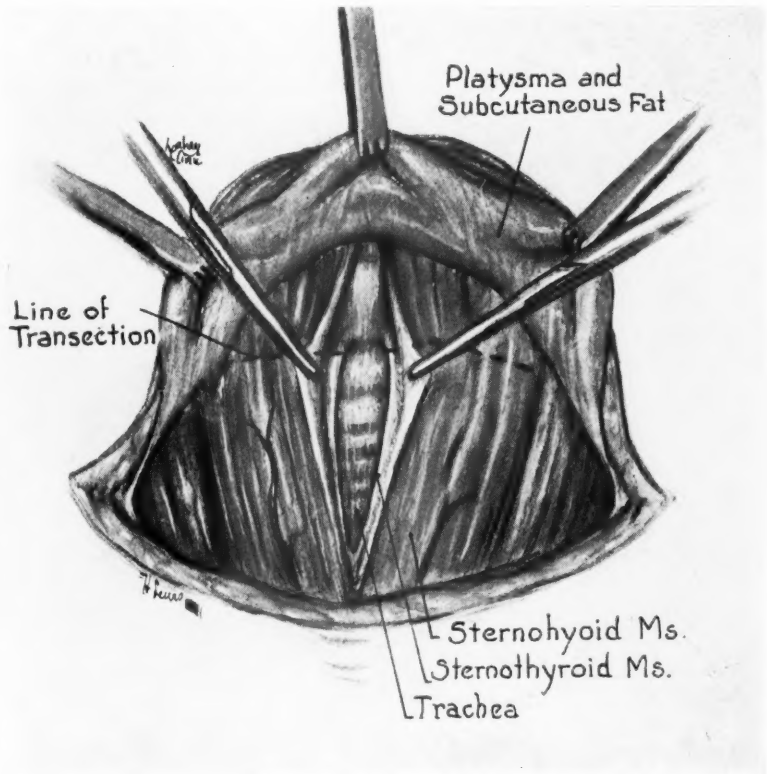


FIG. 11.—The method of folding the skin flap and moving the double hooks from the cut edges down to grasp the double fold of skin flap, thus facilitating its elevation to a high level. The level of transection of the prethyroid muscles is shown, as well as the second layer of muscles, the sternothyroid.

and prethyroid muscles. The flap, then consisting of skin, subcutaneous fat and platysma, can be delicately dissected up until it is well established, and by means of gauze pads can be wiped up with relative bloodlessness to any upper level of height which is desired. This is important because if the incision is carried up between the skin and platysma, the well vascularized subcutaneous fat bleeds considerably and requires considerable time and fussing for control. If the incision is unduly deepened beyond the platysma, the large, prominent veins adherent to the prethyroid muscles will be severed, control of the bleeding will consume considerable time, and the blood will interfere with the anatomic dissection of the skin flap upward.

I do not wish to give the impression that a good goiter incision cannot be obtained by elevating the skin flap in the subcutaneous fat plane between the skin and prethyroid muscles. From my experience, however, there are real disadvantages to the elevation of a skin flap in this plane: (1) Since dissection actually must be upward, it is more difficult and requires a greater amount of time to control the bleeding; and (2) greater postoperative edema occurs since traumatized fat reacts with swelling and edema in the skin

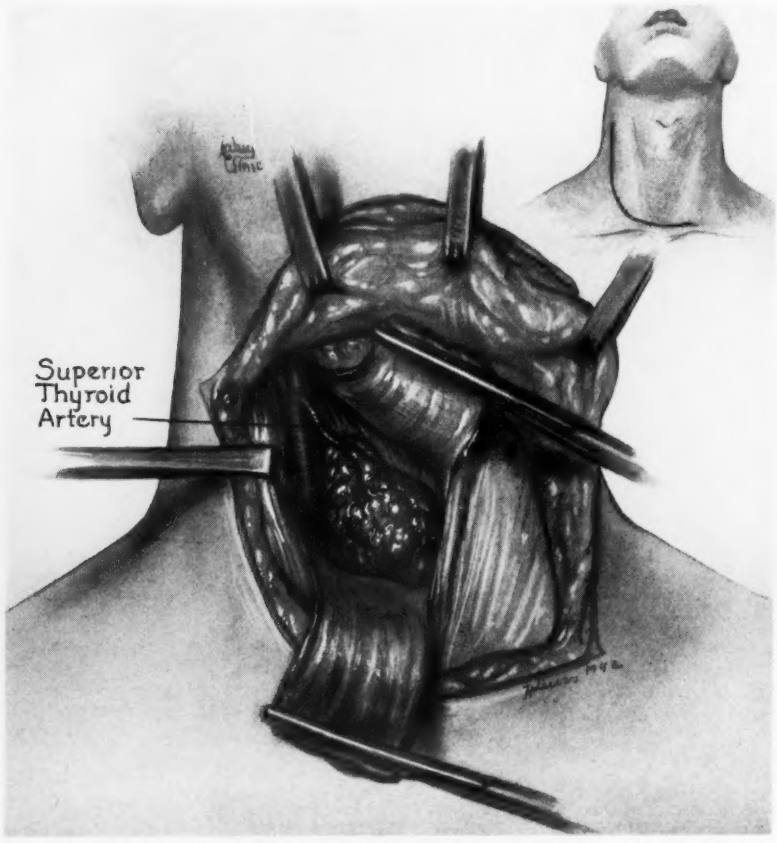


FIG. 12.—The insert shows the method of continuing the goiter incision upward on the side of the neck when serious secondary superior thyroid hemorrhage has occurred postoperatively. The large illustration shows the exposure of the first branch of the external carotid, the superior thyroid. This exposure facilitates prompt and complete control of superior thyroid artery hemorrhage.

flap much more than when the fat layer is retained untraumatized and when the flap is elevated in the plane as described, between platysma and prethyroid muscles. In addition, when the skin flap is elevated in the layer of subcutaneous fat between the skin and prethyroid muscles, dimple-producing, synechia-like scars develop postoperatively between the skin and subcutaneous fat. This does not leave the same freely movable skin flap that results when the subcutaneous fat layer between the skin and platysma remains undisturbed. In addition, the less trauma there is to

the subcutaneous fat in the immediate vicinity of the wound the less reaction there is in the scar. I believe it is of real value, therefore, to grasp the platysma, once it has been demonstrated by the procedure above described (Fig. 10 c), with double hooks throughout the wound so that as the flap is lifted up and wiped up the platysma is not pulled away from the wound edge of the upper skin flap and a large amount of subcutaneous fat exposed.

We have found it unnecessary to undermine or to pull down the lower skin flap. Literally thousands of goiter incisions have been made in this Clinic without mobilizing the lower skin flap in any way and without impairing the appearance of the scar.

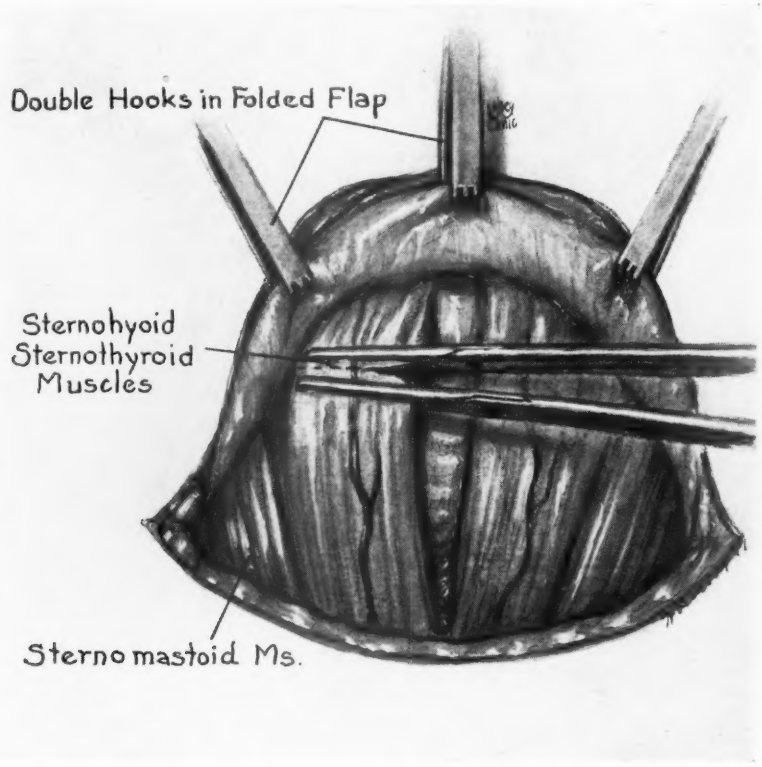


FIG. 13.—The method of grasping the double fold of upper skin flaps to facilitate its upward dissection well above the high level at which the prethyroid muscles are cut in order to avoid destroying their innervation and to stagger the muscle suture and skin suture at different levels (Fig. 15).

As the assistant soon learns, elevation of the skin flap up to the level of the notch of the thyroid cartilage is relatively simple since the fascial plane between the prethyroid muscles and the platysma is loose and well established up to this level. From here on it is not as well established and frequently requires sharp dissection. In order to obtain adequate exposure of the point where the superior thyroid artery enters the upper pole of the thyroid, all skin incisions should be elevated well above the level at which the upper pole of the thyroid can be palpated. This high elevation of the

skin flap often requires considerable determination on the part of the surgeon and willingness to face at times troublesome oozing from small vessels. In my opinion, no goiter incision is satisfactory unless the skin flap is elevated to such a level that at least an inch of the superior thyroid artery can be seen beyond the point at which it enters the apex of the thyroid gland. Figure 11 shows the method of grasping the folds of the upper skin

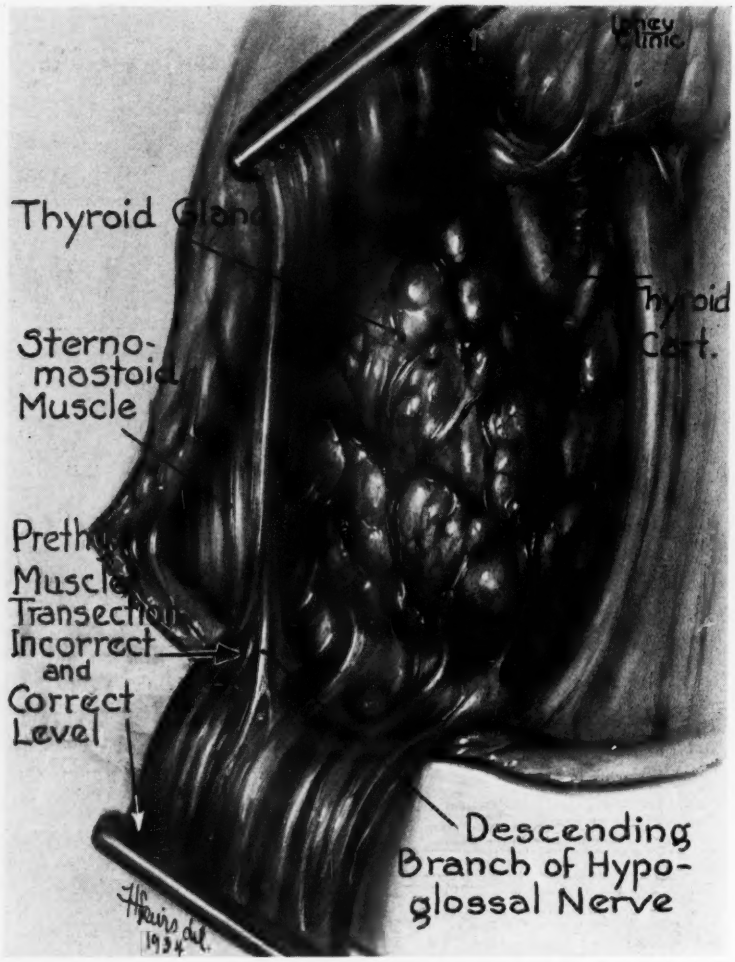


FIG. 14.—Innervation of the prethyroid muscles by the descending branch of the hypoglossal nerve at the point at which it is injured when the incision is incorrectly made at a lower level.

flap in order to get adequate height of exposure. When a skin flap is so elevated, double ligation of the thyroid artery well off the thyroid gland with two ties is possible. This insures against postoperative hemorrhage from the superior thyroid artery, a most troublesome hemorrhage, since when a ligature on the superior thyroid artery slips, the bleeding vessels retract up into the fascial plane of the neck, and it is most difficult to clamp and control them without wide exposure. This clear exposure by high elevation

of the skin flap is important as lack of it, I am certain, has resulted sometimes in inadequate exposure of the upper pole and inaccurate ligature of the superior thyroid artery. A tie about the superior thyroid artery that includes also the tip of the thyroid gland when the skin flap has not been elevated enough to provide a good exposure often shuts off the vessel temporarily but does not permit complete and dependable ligature of the vessel. This frequently permits the patient to cough off the inaccurately applied tie, and results in trying and dangerous postoperative hemorrhage. Such adequate exposure of the superior thyroid vessel with ligation of the superior

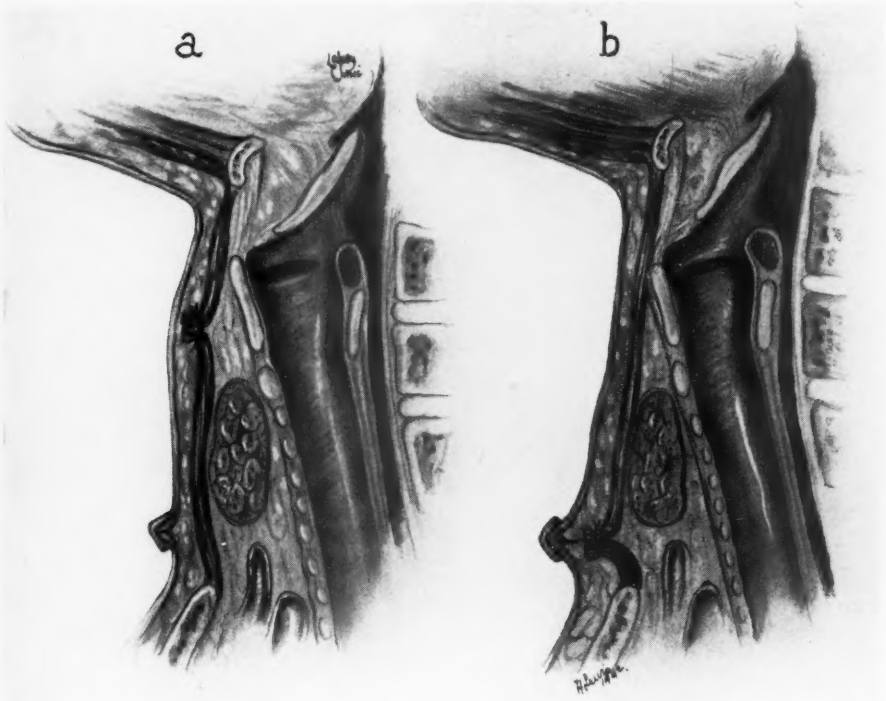


FIG. 15.—(a) A side view of the proper level of transection and suture of the prethyroid muscles. Note the staggering of the high muscle suture and the low skin suture, thus producing no unsightly wound projection or complication. (b) The unfortunate situation which results when the muscles are cut low and muscle suture and skin suture are at the same level.

thyroid vessel well above the tip of the thyroid tissue which forms the upper pole will almost eliminate the possibility of postoperative hemorrhage from the superior thyroid artery.

Should a hemorrhage from the superior thyroid artery occur, unless the bleeding vessel can be seen immediately and plainly on reelevation of the skin flap, one should not waste time in trying to find the vessel in the fascia and muscle plane filled with hematoma. The incision should be extended upward on the side (Fig. 12), the external carotid artery found and the first branch of the superior thyroid artery tied. This procedure, placing, as it does, the patient's life above the beauty of the scar, affords immediate control of

bleeding in a toxic patient sometimes too ill to be submitted to more than one operative procedure for the control of a postoperative hemorrhage.

I know of no surgical procedure which has occasioned greater disagreement and more argument than the question of whether one should cut across the prethyroid muscles, sever them and resuture them, or whether subtotal thyroidectomy should be performed with the prethyroid muscles uncut and retracted to one side. A subtotal thyroidectomy can be undertaken very well without severing muscles, but I personally am not interested in operating upon a patient for thyroid disease without the best possible anatomic exposure of the thyroid and its adjacent and related structures. If an

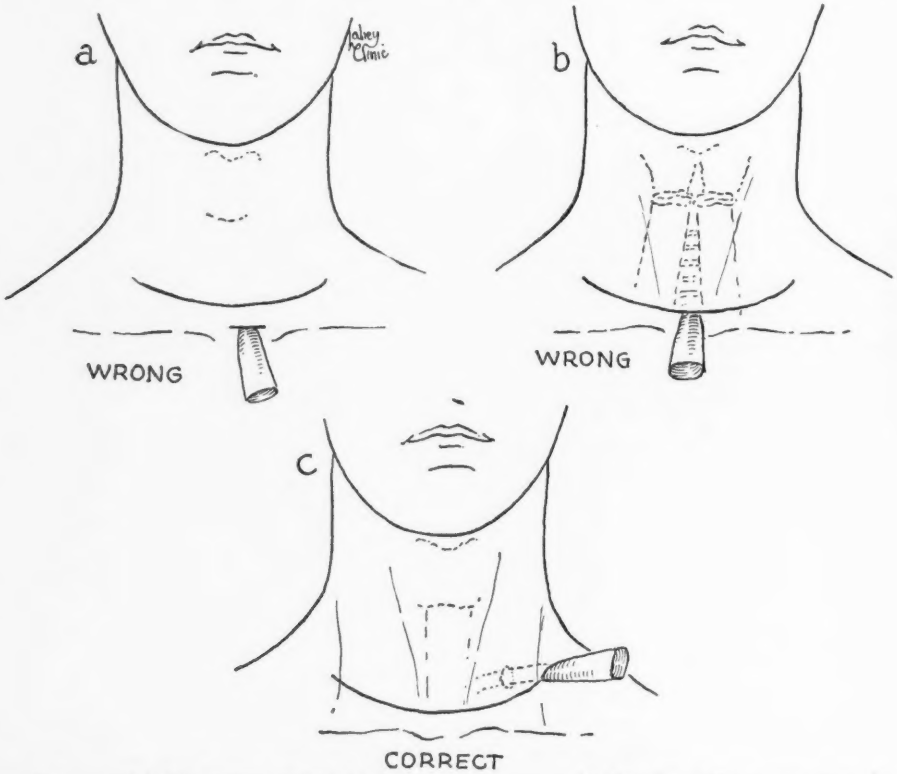


FIG. 16.—(a) The wrong method of drainage. A counter incision for drainage can never be concealed. (b) The drain must pass between the prethyroid muscles when brought out through the central portion of the wound. The skin can become adherent to the trachea when the drain is withdrawn, thus producing upward and downward motion of the scar with swallowing. (c) The proper method of drainage, either through or over the sternomastoid muscle, the drain emerging through the external angle of the wound. With the drain so inserted, the prethyroid muscles can be accurately sutured in the midline, and there are no adhesions between the skin and trachea.

adequate incision is essential in order to obtain exposure for a cholecystectomy, hysterectomy or nephrectomy, it is doubly necessary for a successful subtotal thyroidectomy. Many operations for hyperthyroidism involve risk of the patient's life. Anything which facilitates the exposure adds to the ease of execution of the operation and makes easier the determination of what proportion of the entire thyroid gland is to be removed, and by demonstration of its anatomic relation to the parathyroids and recurrent lary-

geal nerves, adds greatly to the ease and success with which these structures can be preserved. We have demonstrated, conclusively, that if the prethyroid muscles are cut and sutured high there is no functional disability and no disfigurement. If I must choose between cut muscles which can be sutured satisfactorily and a greater hazard of injury to a parathyroid or a recurrent laryngeal nerve with the muscles uncut, I will, and have, consistently chosen the former.

Because we have performed so many operations upon toxic goiters in two stages, we have had the opportunity to cut the muscles on one side, sew

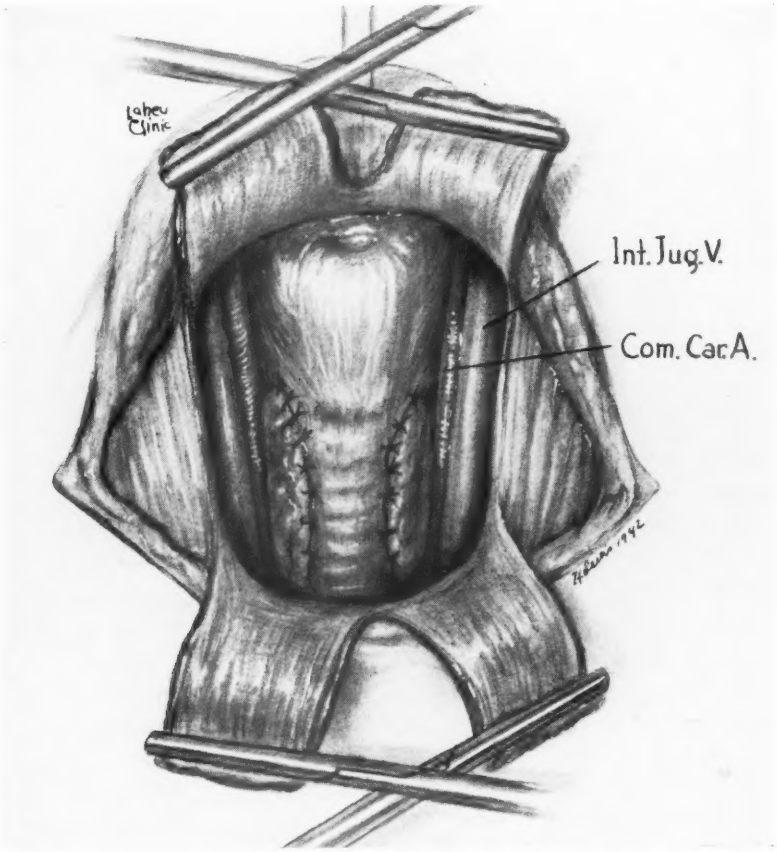


FIG. 17.—The method, which we have repeatedly demonstrated, of suturing the remaining stump of thyroid against the trachea to overcome oozing and thus permit closure of the wound without drainage.

them together, have the patient back at the end of six weeks for the second stage (left subtotal hemithyroidectomy) and observed the condition of the muscles. Invariably one can hardly find the place where the muscles have been sutured, if the incision is made high so that innervation is not disturbed and there is no disfigurement or disability (Fig. 13). The two essential features involved in transection of the prethyroid muscles, in order to obtain better exposure, are that they must be incised high and sutured high,

well up under the upper portion of the wound (Figs. 13 and 14). This avoids denervation of the prethyroid muscles, and, most importantly, staggers the muscle sutures high up under the skin flap (Fig. 15 a) and not low on the same level with the skin incision (Fig. 15 b). The easiest level at which to transect the prethyroid muscles is low, where the nerve innervation is endangered, and where the muscle suture rests close to, or immediately beneath, the skin incision (Fig. 15 b). This error tends to produce unfortunate wound complications and to make undue prominence from the double line of sutures of skin and muscle directly beneath the skin incision.

I am sure that high severing of the prethyroid muscles, clamped well out beneath the sternomastoid, which has been freed and pulled back, has resulted in infinitely better exposure, in drier fields, more anatomic dissections, and more radical dissection, and has resulted in neither organic dysfunction nor disfigurement.

In closure of the skin wound the prethyroid muscles must be sutured accurately and without tension. The muscles in the midline are best brought together with interrupted sutures, making certain they are accurately approximated between the skin flap and the trachea, thus serving to prevent the trachea from becoming adherent to the skin.

If drains are inserted, no counterincision should be made beneath the skin incision through which the drain is to emerge (Fig. 16 a) because this produces an ugly scar which cannot be covered by a necklace. Likewise, if a drain is inserted, and they are rarely employed at this Clinic, it should be brought out at the external angle of the wound either through the belly of the sternomastoid (Fig. 16 c) or in front of the sternomastoid between it and the sutured prethyroid muscles. This brings the drain out through the corner of the incision where the skin incision cannot become adherent to the trachea. If the drain is brought out in the midline between the prethyroid muscles (Fig. 16 b), these muscles gap and the organized scar which fills the hole between the prethyroid muscles when the drain is withdrawn, permits the trachea to become adherent to the skin, making the scar "bob" up and down when the patient swallows. It is well to bear in mind, however, that if bleeding is painstakingly controlled and if the cut surface of the thyroid is turned against the trachea and sutured there, as we have repeatedly advised (Fig. 17), rarely, except in the large intrathoracic goiters is it necessary to use drains. With wound closure without drainage we have had no more accumulation of serum in the wound than when drains were employed.

Finally, for many years, in closure of the skin we have employed no subcutaneous sutures in the platysma. Omitting the suturing of the platysma muscle in no way results in spreading of the scar, as we have also proved by experience with many patients in whom no platysma sutures have been employed.

The skin, closed as it is without drainage, is approximated accurately with clips. The care taken in accurately placing cut skin edges together with clips plays a great part in the appearance of the scar. If the upper

edge of the cut skin is permitted to overlap the lower or the cut edges are at all inaccurately approximated, definite thickening of the scar will result. Clips are put fairly close together to control skin oozing, and most importantly, one-half of the clips are removed on the second day and the other half on the third. As a matter of fact, if one wishes, they can all be removed on the second day. There is not the slightest danger of the patient pulling the skin edges apart, because if he elevates the chin he pulls on the sutured prethyroid muscles and does not exert pressure on the skin wound. If skin clips are left on for longer than three days, pressure necrotic spots will develop and result in white dot scars, thus permanently disfiguring the wound.

CONCLUSIONS

Deductions drawn from over 22,000 goiter incisions are presented. In my opinion, however illogical it seems, and even though the physical improvement may be striking, nothing plays a greater part in the happiness of a patient operated upon for goiter than the postoperative appearance of the incision. Technical points which I have found useful in making better goiter incisions and obtaining better scars are discussed.

THE PRE- AND POSTOPERATIVE USE OF A METAL-TIPPED GASTRODUODENAL TUBE AS AN AID IN THE SURGICAL TREATMENT OF DUODENAL OBSTRUCTION IN THE NEWBORN

E. P. Maris, M.D., A. C. McGuinness, M.D., H. F. Lee, M.D., Jonathan E. Rhoads, M.D., and Walter Estell Lee, M.D.

Philadelphia, Pa.

FROM THE CHILDREN'S AND THE PENNSYLVANIA HOSPITALS OF PHILADELPHIA, AND THE DEPARTMENT OF PEDIATRICS, SCHOOL OF MEDICINE, UNIVERSITY OF PENNSYLVANIA

Congenital obstructions of the duodenum have always presented a difficult surgical problem, and the mortality has remained high in spite of numerous changes in treatment. Ladd has made an important contribution to the field by showing that most of those obstructions due to extrinsic causes are associated with faulty rotation of the large bowel with or without volvulus of the midgut.

It is evident, in reviewing statistics on this subject condensed from Ladd and Gross[1] (Table I), that the mortality of intrinsic obstruction has remained much higher than that experienced from extrinsic obstruction since the introduction of Ladd's operation for the latter. It also seems evident, from the figures cited, that anastomosis is extremely hazardous in the infant and that the lower in the bowel anastomosis is performed the greater the danger. Whether the high mortality associated with simple enterostomy is due to the danger of opening the bowel or whether it is due to the great difficulty of supportive treatment after these procedures, is not clear from the statistics. One of us (W. E. L.) has performed jejunostomy with success in two newborn infants.

It seems possible, therefore, that a small opening in the bowel is not as dangerous as has sometimes been believed, but that the mortality associated with enterostomy has been due to metabolic disturbances and to the selection of patients with poor vitality for this procedure.

Table I

MORTALITY IN CONGENITAL STENOSIS AND ATRESIA OF THE SMALL BOWEL, COMPARED WITH THE MORTALITY IN CONGENITAL OBSTRUCTION OF THE DUODENUM DUE TO EXTRINSIC CAUSES

Condensed from Ladd and Gross

	Jejunostomy or Ileostomy		Gastro-enterostomy		Entero-enterostomy	
	No. of Cases	Mortality Per Cent	No. of Cases	Mortality Per Cent	No. of Cases	Mortality Per Cent
Stenosis of duodenum	1	100%	2	50%	7	72%
Atresia of duodenum	0	—	1	—	4	100%
Stenosis of jejunum	0	—	2	50%	—	—
Atresia of jejunum	0	—	2	—	6	50%
Stenosis of ileum	3	100%	0	—	4	50%
Atresia of ileum	22	100%	0	—	12	75%
Total (intrinsic obstruction)	26	100%	7	50%	33	70%

Total of all intrinsic obstruction.....64 cases — 16 lived, or 75 per cent mortality.
Total of all extrinsic obstruction.....44 cases — 27 lived, or 38 per cent mortality.
Total of extrinsic cases treated by Ladd's method...35 cases — 27 lived, or 23 per cent mortality.

A difficulty which we believe may contribute to the mortality in this group of patients is that of being sure that no intrinsic obstruction remains after adhesions have been released. The duodenum is to some extent bound down normally, and when it becomes dilated its fibrous attachments become taut and compress it. When these attachments are divided, the bowel seems to be released and the area of dilatation may even appear to advance slightly.

There is another situation which may lead to the termination of an operation before the lumen of the duodenum is clear. It is possible for the small bowel to be obstructed by two or more diaphragms (Fig. 1). After the release of the first obstruction bowel contents start to pass but may not be followed far enough to reveal the next obstruction.

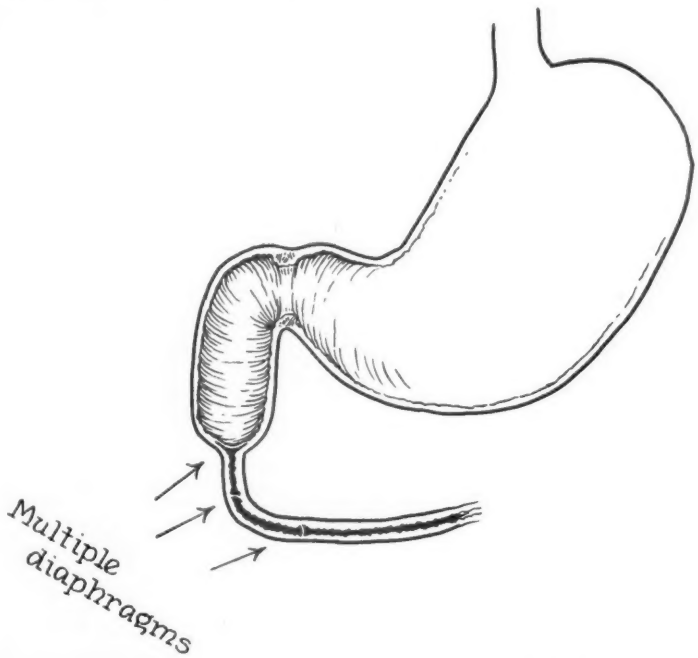


FIG. 1.—Diagram showing duodenal obstruction with multiple diaphragms.

Therefore, we have found it to be a useful maneuver to introduce a special tube into the duodenum before operation, which has a metal tip large enough to be grasped and manipulated through the bowel walls. The stomach is emptied *en route*. In cases of duodenal obstruction all possible extrinsic causes should be excluded by freeing the duodenum throughout its length, after the right side of the colon has been reflected to the midline or beyond, taking care to avoid injury to the mesenteric vessels, when it will be possible to free the duodenojejunal flexure so that the duodenum is relatively mobile for its full length from the pylorus to the ligament of Treitz.

The tip of the tube is then manipulated as far as the proximal jejunum. If any intrinsic obstruction is present, it should be readily detected. In the event that the obstruction is due to a diaphragm, a small opening can be made

with a transverse incision through the antimesenteric border of the bowel, at a point one to two inches below the obstruction, through which a hemostat, can be inserted as in Fig. 2 to grasp the metal tip of the tube (and the diaphragm) and thus tear or bite an opening through the diaphragm and draw the tube four or more inches into the lumen of the bowel distal to the obstruction. Subsequently, the incision in the bowel wall is closed transversely with two layers of sutures.

The following four case reports illustrate the use of such tubes:

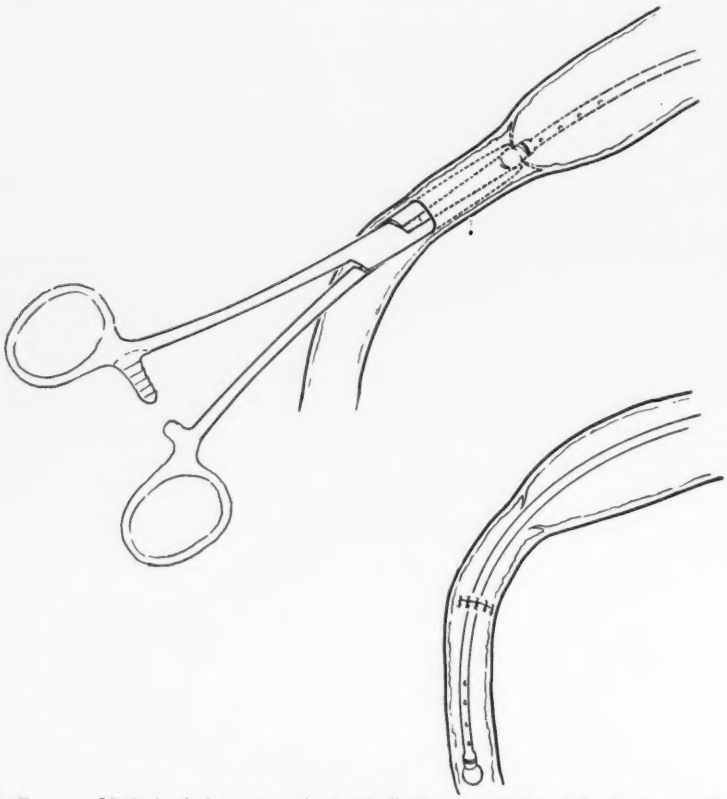


FIG. 2.—Method of destroying duodenal diaphragms employed in Cases 1 and 4. The only opening in the bowel is small, antimesenteric, and is placed below the obstruction through normal bowel wall. It is closed transversely.

ILLUSTRATIVE CASES

Case 1.—Baby S. T. A., a female infant, regurgitated bile-stained material from birth. Roentgenologic examination, at the age of four days, showed dilatation of the stomach and duodenum but practically no air in the remainder of the intestinal tract (Fig. 3). An operation under ether anesthesia revealed numerous tight fibrous bands across a distended duodenum. These were released. The collapsed bowel did not fill-out much but it was thought that it did increase in diameter slightly. The condition of the child being critical, the abdomen was closed.

Vomiting continued, and roentgenologic examination, made three days after operation, again showed an absence of air in the intestine. A brass-tipped gastroduodenal tube (No. 10 F.) was then manipulated as far as the duodenum under fluoroscopic guidance and the wound reopened. An attempt to pass the tube along the duodenum revealed, by palpation and visualization of the brass tip, the presence and location of a

diaphragm. This was then destroyed, as shown in Figure 2, and the tube drawn about three inches into the lumen of the jejunum. Following this operation the baby began to have yellow stools and gradually to gain weight. Roentgenologic examination a few days later showed a considerable quantity of gas in the intestine (Fig. 4). The patient

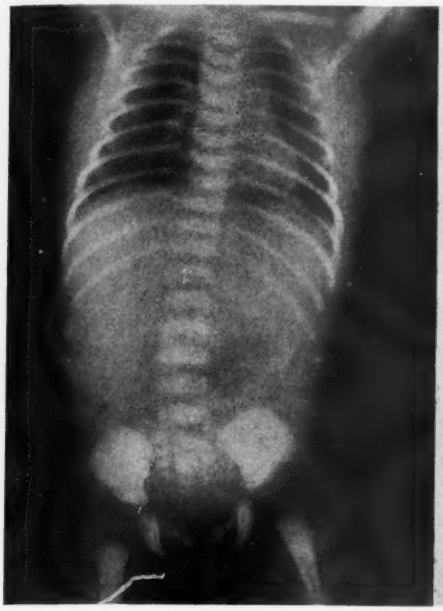


FIG. 3.—Case 1: Roentgenogram of Baby S. T. A. before relief of the obstruction.

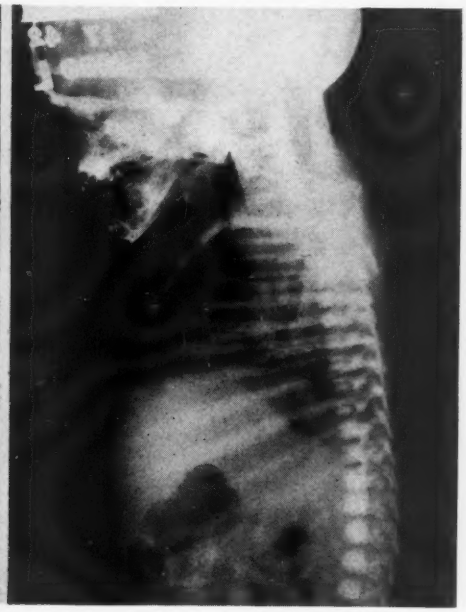


FIG. 4.—Case 1: Roentgenogram of Baby S. T. A. after relief of the obstruction.

was discharged from the hospital, in good condition, four weeks later, and reached normal weight at the end of the third month.

The conditions encountered in Case 1 are shown diagrammatically in Figure 5.

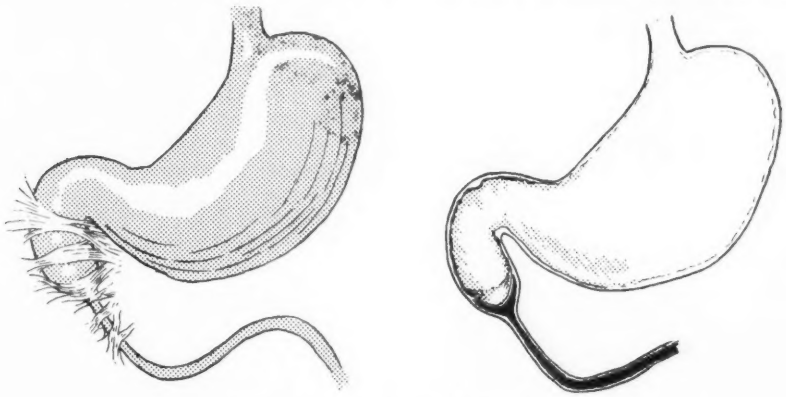


FIG. 5.—Diagram of the operative findings in Case 1.

Case 2.—Baby S. T. I., a male infant, vomited greenish material from birth. Roentgenologic examination, with barium, showed duodenal obstruction. Prior to operation the brass-tipped gastroduodenal tube was passed. In this infant adhesions over the second portion of the duodenum had to be divided. It was again found difficult to pass the tube into the distal duodenum but it finally slipped through a strictured area, and some three inches of the tube was left in the proximal jejunum. Subsequently, six hours later, the tube had to be removed because of gagging and respiratory em-

barrassment, but, nevertheless, the child began to pass stools within 24 hours and he has gradually gained weight and strength, and his bowels were functioning normally three weeks postoperatively. At six months of age he weighs 17 pounds, has the usual diet and has reached the normal level of development.

Case 3.—Baby A. G., a female child, was admitted to the Children's Hospital because of persistent vomiting of bile-stained material. Operation was deferred until the 27th day of life because the obstruction was not complete. After conservative treatment during this period had failed to result in improvement, operation was undertaken,

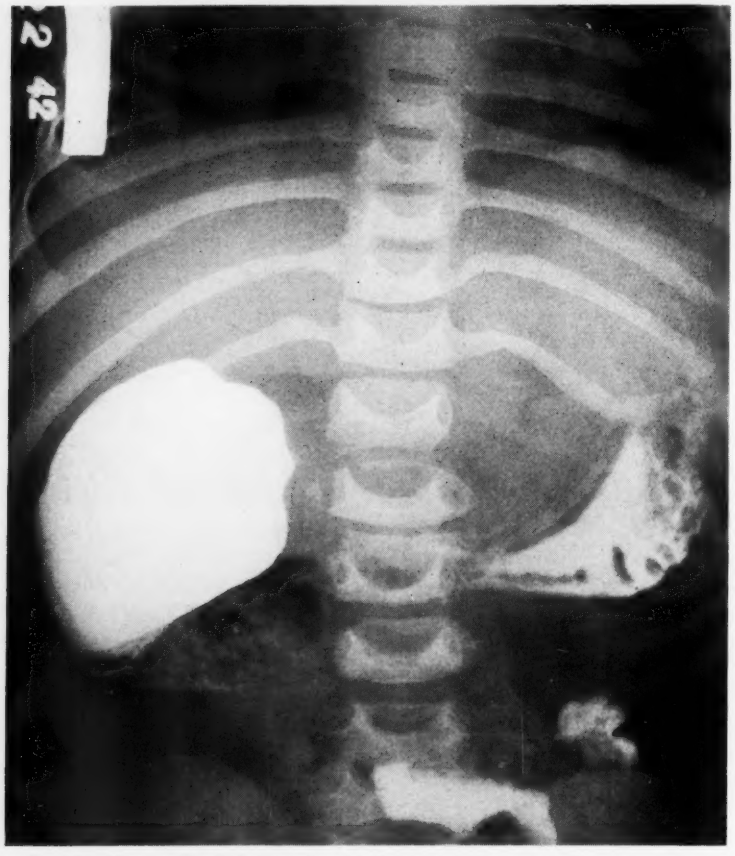


FIG. 6.—Case 4: Marked dilatation of the duodenum at the age of two years.

with the aid of a No. 10 F. brass-tipped duodenal tube. The duodenum was found to be dilated and rather tight bands passed over it near the duodenojejunal flexure. After the right half of the colon was reflected to the midline, these bands were divided. It was then found that some of the small intestine had become adherent on the lateral side of the mesentery of the ascending colon. The brass-tipped tube was of decided value in determining the patency of the duodenum in the confusing anatomic anomalies found at operation.

After freeing all of the abnormal bowel attachments, the abdomen was closed and, except for respiratory difficulty necessitating removal of the tube during the period of reaction from anesthesia, a satisfactory convalescence followed.

Case 4.—Baby R. J. L., a two-year-old female child, was admitted to the Children's Hospital on account of recurrent episodes of vomiting. The vomitus was usually bile-stained. Roentgenologic examination showed evidence of partial obstruction in the descending portion of the duodenum, with an extreme degree of dilatation (Fig. 6).

At operation, the duodenum was exposed by reflection of the colon and a No. 12 F. Miller-Abbott tube, which had been previously introduced, was moved as far down the duodenum as possible. In the last portion of the duodenum the tip was deflected toward the mesenteric side by a shelf of tough fibrous tissue. Here it caught in a pocket and could not be advanced further.

The bowel was opened below the obstruction and along its antimesenteric border, as in Case 1, and the tip of the tube, together with the walls of the diaphragm, were seized with a hemostat. As they were drawn down and through the incision in the duodenal wall the diaphragm was incised, and the brass tip and tube drawn into and left several inches in the lumen of the jejunum. The opening in the bowel was closed transversely.

Convalescence was satisfactory, and the patient's symptoms were relieved immediately. In this case, had it not been for the tube, an anastomosis probably would have been necessary. With this patient the tube remained in the jejunum for three weeks, and was used for feeding.

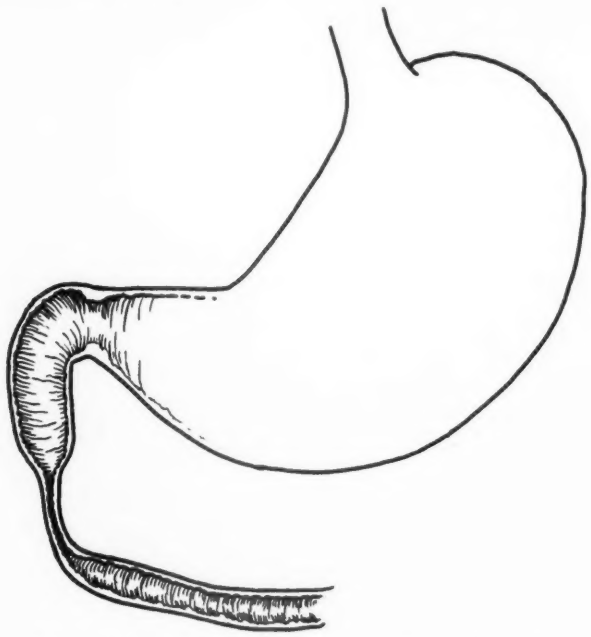


FIG. 7.—Narrowing of the duodenum without complete obstruction. This condition was found in Case 2.

DISCUSSION.—In attacking the problem of congenital duodenal obstruction, it is desirable to avoid anastomosis and to open the bowel as infrequently as possible. When it is necessary to open it, the incision should be as short as possible, and should preferably be made through normal bowel wall at a point where it will not interfere with the blood supply of adjacent portions.

A duodenal tube, such as is shown in the insert in Figure 2, may perform four functions. It is employed, first, to deflate the stomach. Next, when followed fluoroscopically, it may give additional evidence on the point of the obstruction. Third, it is available at the time of operation to test the patency of the duodenum after the latter has been freed. Fourth, it makes possible the destruction of a diaphragm, by the method illustrated in Figure 2.

This method fulfills the criteria outlined above rather well, and should entail a relatively small risk of peritonitis.

A word of caution is necessary in regard to the employment of the gastroduodenal tube during the early postoperative period. The two-year-old child was not disturbed by the presence of the tube at this stage. All three of the newborn infants, however, each of whom tolerated the tube well preoperatively and during the operation, showed evidence of serious respiratory difficulty, and a condition resembling shock at some time during the period of reaction from anesthesia. Prompt removal of the tube resulted in relief in each instance. From our experience, it would seem advisable to remove the tube before complete reaction from the anesthesia takes place. Eighteen to 24 hours postoperatively the tube has been reinserted, for relief of distension, without the recurrence of unfavorable signs.

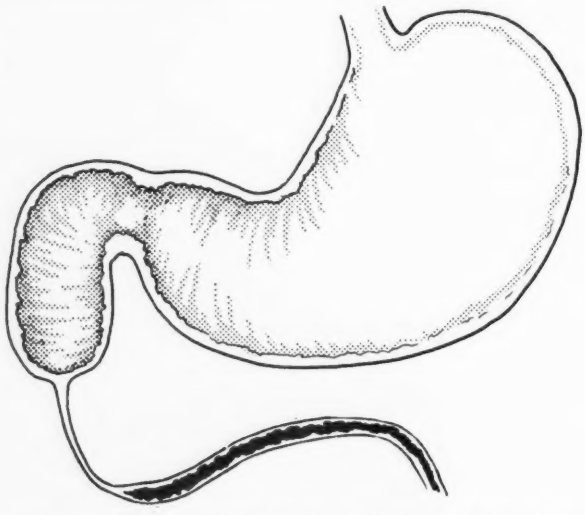


FIG. 8.—Complete atresia of the duodenum requiring gastro-jejunal anastomosis.

If no diaphragm is present and only a stenosis is found, it may be possible to pass the tube through the narrowed area, as in Figure 7. In certain cases an atresia of a considerable segment of bowel is present (Fig. 8). In these cases there would seem to be no alternative other than to perform an anastomosis.

CONCLUSION

A duodenal tube with a metal tip of sufficient diameter to be visualized and grasped through the bowel, is recommended as an adjunct in operations for relief of congenital duodenal obstructions.

Its use in four cases is described which are illustrative of its value.

We wish to acknowledge our indebtedness to Dr. Paul Bishop and Dr. Ralph Bromer for the roentgenograms, and to the Harrison Department of Surgical Research, University of Pennsylvania, for assistance with the illustrations.

REFERENCE

1 Ladd, W. E., and Gross, R. E.: Abdominal Surgery of Infancy and Childhood. W. B. Saunders, Philadelphia, 1941.

CONGENITAL CYSTIC DILATATION OF THE COMMON BILE DUCT*

CASE REPORT AND REVIEW OF LITERATURE†

THOMAS A. SHALLOW, M.D., SHERMAN A. EGER, M.D.,
AND
FREDERICK B. WAGNER, JR., M.D.
PHILADELPHIA, PA.

THE UNIQUE ASPECTS of congenital cystic dilatation of the common bile duct, otherwise known as choledochus cyst, make it interesting, notwithstanding its rarity. The relatively few reports in the literature warrant the accumulation of all the available data in order to arrive at a thorough understanding of this subject. With this in mind a complete review of the literature is made and a case report is included.

The outstanding features of the case, herewith cited, are as follows: The condition was suspected preoperatively, and peritoneoscopy was undertaken as an adjunct in diagnosis. The cyst was extirpated and right and left hepaticoduodenostomy performed over No. 18 F. catheters. The latter were removed through a gastroscope by Dr. Clerf during a second hospital admission, 11 weeks postoperatively. Recovery ensued.

Case Report.—P. W., white, male, age 20, was admitted to the ward medical service of the Jefferson Medical College Hospital July 11, 1941. The chief complaints were pain in the right upper abdomen, a mass in the same area, jaundice, and fever.

The patient had been well until November, 1940, at which time he experienced his first attack of pain in the right upper abdomen. The pain was definitely localized at the right rectus-costal margin junction; it was sharp, did not radiate, and lasted for three to four days, with acute exacerbations seven or eight hours apart. This pain was followed within 24 hours by a mass in the same area, jaundice, and slight fever. He was studied for one week at his College Infirmary and discharged to the care of his family physician, with a diagnosis of catarrhal jaundice. During the following month the jaundice and fever gradually disappeared, but weakness persisted, and he was advised to remain out of college in order to regain his strength.

From December, 1940 to June, 1941 the patient remained at home and performed small chores. The mass in the right upper abdomen persisted, but without pain, jaundice, or fever. He ate an ordinary diet and took no drugs.

About two weeks prior to admission, the patient again experienced pain in the right upper abdomen, which had the same characteristics previously noted; it lasted for eight hours and was severe enough to force him to bed. The pain was again followed, as in the previous attack, by jaundice and fever. With each attack of jaundice the urine became darker than usual, and the stools were of a light brown color, but not clay-colored. There was a loss of seven to eight pounds in weight with each attack, but this was subsequently regained, and the weight on admission was that normally noted. Vomiting did not occur at any time. A review of all the remaining systems revealed a normal history. His past medical history was uneventful, and the family history failed to reveal any member ever suffering from similar complaints.

---

* Presented before the Philadelphia Academy of Surgery, December 7, 1942.

† From the Samuel D. Gross Surgical Division of the Jefferson Medical College Hospital.

Physical Examination.—The patient was a well-developed, slightly undernourished white male, who appeared chronically ill. Temperature 99.4° F.; pulse 76; respirations 20. Blood pressure 112/70. Weight 143 pounds. He had slight pallor and a greenish tinge to the skin. The scleras were markedly jaundiced. The tongue was coated and a few tonsillar tabs were present on the left side. There was slight anterior cervical lymph node enlargement. The heart and lungs were essentially normal. There was prominence of the superficial veins over the right lateral chest wall. A large, globular mass, about six inches in diameter, was visible in the right upper quadrant of the abdomen, which moved on respiration toward the midline and very slightly downward. It was firm and smooth on palpation, and no definite edge could

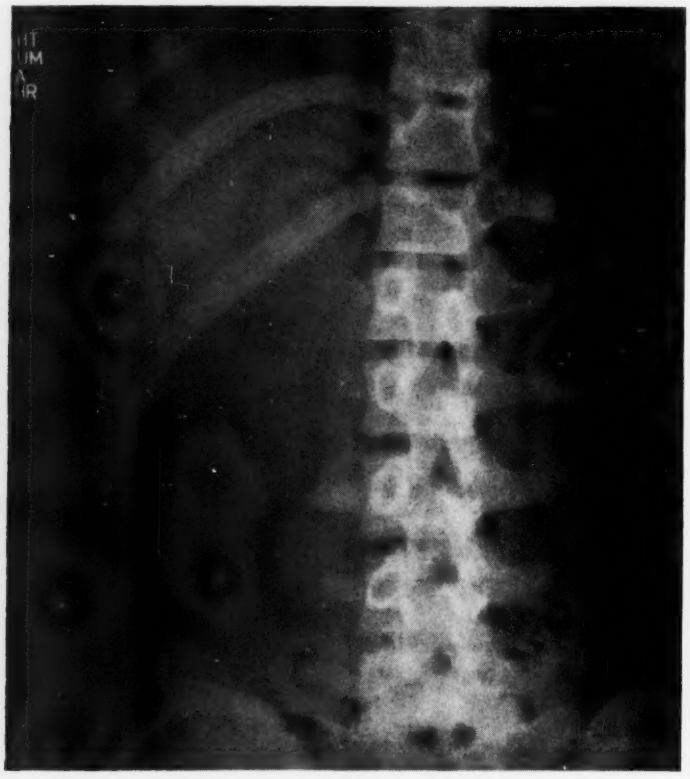


FIG. 1.—Plain film of the abdomen showing the faint outline of the mass.

be made out. There was a resistance to the palpating hand from the right anterior superior iliac spine across the navel to the left costal margin, which was suggestive of hepatic enlargement; percussion note over this area was flat. The remainder of the physical examination was essentially normal. *Diagnostic Impression:* Abdominal mass with obstructive jaundice.

Laboratory Data and Special Studies.—Examination of the blood showed: Hemoglobin 65%; red cells, 3,720,000; white cells, 5,600, with polymorphonuclear cells 48%, lymphocytes 46%, and monocytes 6%. Color index 0.88.

Urinalysis, repeated at intervals of several days, showed deep green color (grossly bile-stained); reaction sometimes acid, sometimes alkaline; specific gravity ranging between 1.009–1.027; albumin varying from none to moderate amounts; sugar, negative; occasional triple phosphate crystals; no red blood cells; pus cells absent in some specimens, and present up to 30 per low power field in others; no casts. Test for presence

of bile pigment was strongly positive in three different specimens. Test for urobilinogen was positive in three different specimens in dilutions varying from 1–5 to 1–40.

Stool examination revealed a normal amount of bile pigment present.

Wassermann and Kahn serologic studies were negative.

The sedimentation rate was rapid, with a fall to 31 mm. within one hour.

Blood sugar (fasting) was 77 mg. Glucose tolerance test revealed a fasting sugar of 72; ½ hr. 128; 1 hr. 113; 2 hrs. 63.

Serum cholesterol was 163 mg. and one week later 173 mg.

Serum phosphorus was 3.6 mg. and serum phosphatase 21.4 Bodansky units.

Serum proteins were 6.23 Gm., with albumin 3.46 Gm. and globulin 2.77 Gm.

Prothrombin times taken several days apart and during treatment were 38%, 40%, 63%, 87%, and 66%, respectively.

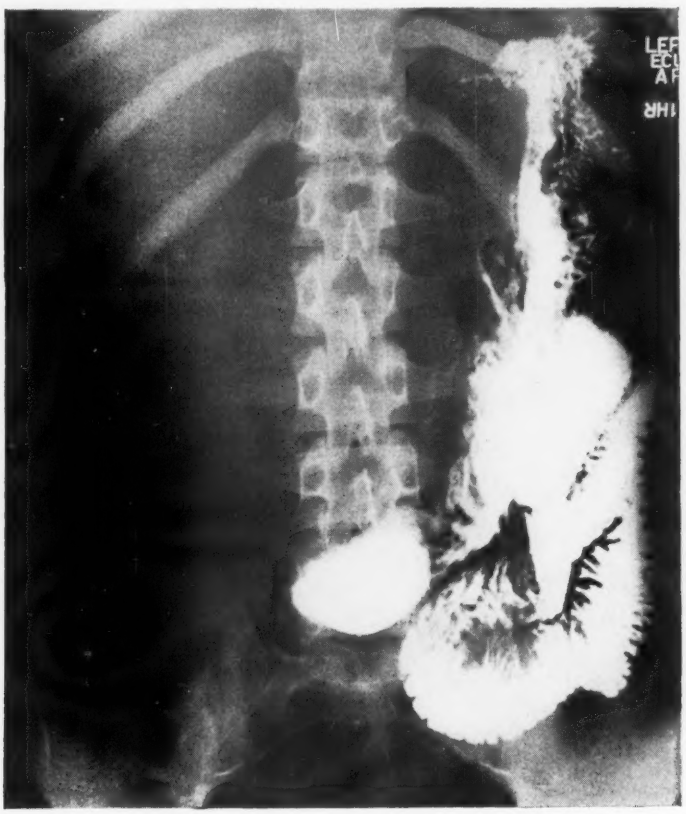


FIG. 2.—Gastro-intestinal series showing displacement of stomach and jejunum.

Bromsulfalein test showed 15% of dye retained, and the test repeated one week later showed 20% dye retained.

Van den Bergh test showed a positive direct reaction, with serum bilirubin of 3.5 mg. The same test repeated one week later again showed a positive direct reaction, with serum bilirubin of 4.6 mg.

Hippuric acid test showed an excretion of 1.20 Gm. as benzoic acid.

Urea clearance was 85% of average normal.

Circulation time was 15 seconds; venous pressure 100 mm. of water; and vital capacity 4,500 cc.

Biliary drainage revealed occasional cholesterol crystals in the A and B specimens, and none in the C specimen.

Sigmoidoscopic examination was normal.

Roentgenologic Studies.—Plain film of the abdomen (Fig. 1): There is a diffuse haze over the upper abdomen which seems to be due to a greatly enlarged liver. The spleen is not visible and both kidneys are normal in size and shape. *Roentgenologic Interpretation:* Enlarged liver.

Cholecystogram: Examination of the abdomen 15 hours after the ingestion of the dye does not show any evidence of a gallbladder shadow. There is a large mass in the right side of the abdomen, a portion of which is enlarged liver. This extends downward as far as the upper portion of the ilium, and it may be that there is a

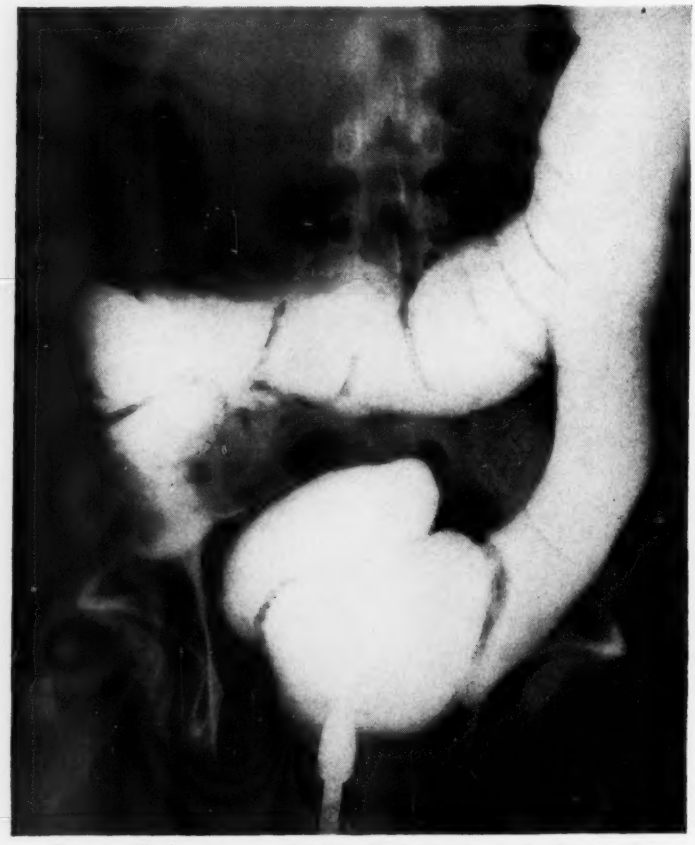


FIG. 3.—Barium enema showing displacement of the hepatic flexure and transverse colon.

mass which is distinct from the liver. A roentgenogram made three hours later still shows no evidence of dye in the gallbladder. *Interpretation:* Nonvisualization of the gallbladder by oral cholecystography. Large mass on right side of abdomen.

Gastro-intestinal Series (Fig. 2): Fluoroscopy of the chest was negative. The esophagus was negative to barium liquid and paste. The stomach was elongated and markedly displaced to the left. Its greater curvature was one centimeter below the iliac crest. The stomach exhibited smooth indentation along its lesser curvature, apparently due to extrinsic pressure. The rugal pattern was intact. The antrum was flattened and displaced anteriorly and to the left, apparently by extrinsic pressure. There was no evidence of an intrinsic organic lesion of the stomach. The duodenum

was displaced downward and markedly anteriorly, especially in the descending limb. Progress through the descending limb was delayed, apparently secondary to extrinsic pressure. These displacements are apparently secondary to a retroperitoneal mass. It may be a retroperitoneal sarcoma or possibly may arise from the pancreas and represent a pancreatic cyst.

Progress through the small intestine was unretarded. The jejunum, however, was displaced to the left and downward. At one hour the barium was distributed through the stomach, duodenum and jejunum. At two hours and thirty minutes the stomach and duodenum were empty and the barium was distributed through the jejunum and ileum. At four hours and thirty minutes the barium had reached the descending colon. There was noted a displacement downward of the hepatic flexure and the transverse colon. The terminal ileum appeared negative. There was a sug-

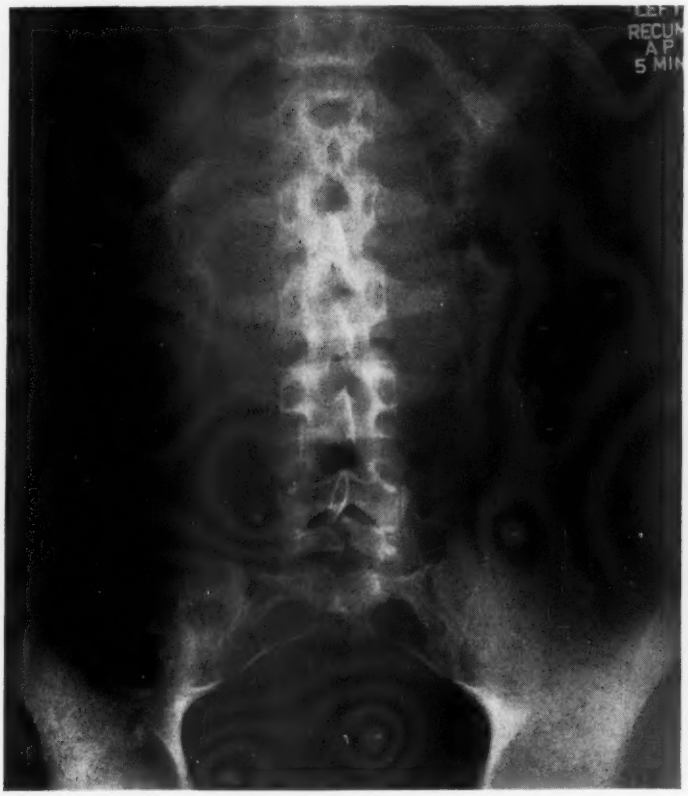


FIG. 4.—Intravenous urogram showing retention of dye in the right renal pelvis, due to pressure on the ureter.

gestion of an incompletely filled appendix. *Interpretation:* Examination of the upper gastro-intestinal tract reveals evidence of extrinsic pressure on the stomach, duodenum, jejunum and colon, apparently due to a right-sided retroperitoneal mass. This may be either a retroperitoneal sarcoma or a pancreatic cyst.

Barium Enema (Fig. 3) : The colon was studied following barium injection. The barium flowed readily throughout as far as the cecum. This did not fill readily. The colon was large in size and was displaced downward in the region of the hepatic flexure, apparently by the previously noted extrinsic mass. Except for the cecal region it filled out readily and exhibited no evidence of an organic lesion. However, the cecal area was noted to fill intermittently and when filled, exhibited no evidence

of a defect. Following evacuation there was noted a small residue in the sigmoid and in the cecal region. *Interpretation:* Barium enema study of the colon shows evidence of extrinsic pressure on the hepatic flexure and possibly on the transverse colon. The cecum filled with difficulty; however, no lesion could be demonstrated.

Intravenous Urogram (Fig. 4): Both kidneys are normal in size, shape, and position. The contrast material was normally excreted by both kidneys, and the pelves and calices have a normal appearance. There is a tendency to retention of the contrast material in the upper part of the right ureter. This might be an expression of early pressure by the abdominal mass that is present. However, one hour after injection, practically all of the contrast material had been eliminated from the kidneys. The bladder has a good degree of density and is small in outline. *Interpretation:* Normal appearing and functioning kidneys. The abdominal mass might be beginning to press upon the right ureter.

Peritoneoscopic Examination: This was performed under local 1% novocain anesthesia. The peritoneal areas were well visualized. The only abnormal finding was a

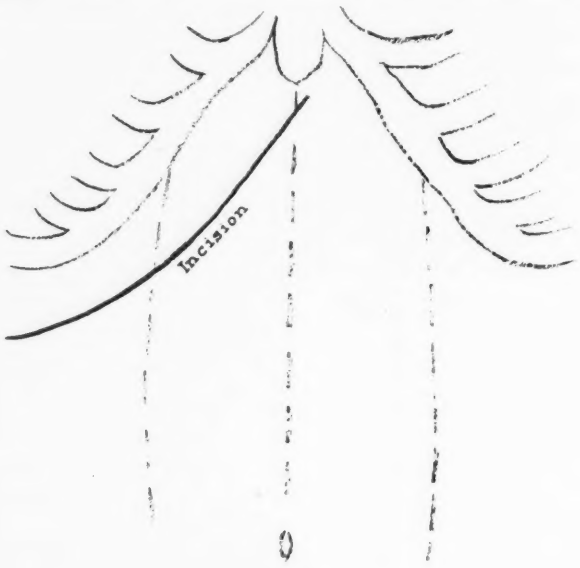


FIG. 5.—Operative approach to choledochus cyst.

large retroperitoneal mass which displaced the duodenum anteriorly and medially, the liver superiorly, and the hepatic flexure of the colon inferiorly. The mass was circumscribed and was soft, probably of cystic nature. *Impression:* Retroperitoneal mass near the right kidney of cystic nature, possibly a cyst or a polycystic right kidney.

Preoperative Course.—The patient was given a high caloric, high carbohydrate, moderate protein, low fat, high vitamin diet. Because of the low prothrombin time, vitamin K was administered intravenously daily, and with this therapy the prothrombin time approached more nearly the normal levels (see laboratory data). Sodium phosphate was also administered orally each morning. During this time the patient was being studied carefully, the results of which are given under the laboratory data and special studies. A low-grade fever was present, there being usually a slight rise daily from normal to seldom more than 100° F. On July 29, 1941, the patient was transferred to Men's Surgical "A" Service. *Final Preoperative Diagnosis:* Retroperitoneal mass, probably choledochus cyst, with obstructive jaundice.

Operative Procedure: Excision of cystic mass, gallbladder, and cystic duct; implantation of the right and left hepatic ducts into the first portion of the duodenum over No. 18 F. catheters.

Operation.—July 31, 1941: The patient was given nembutal gr. 1½ at 7 A.M. and 9 A.M., and morphine sulfate gr. ¼ at 1 P.M. At 1:30 P.M. pontocaine (16 mg.) spinal anesthesia was administered. Abdomen was then prepared with iodine and alcohol and draped. An incision (Fig. 5) was made one inch below and parallel to the right costal margin, and the peritoneal cavity opened. A large retroperitoneal mass (Fig. 6) was seen in the right upper abdomen, displacing the duodenum medially and anteriorly,

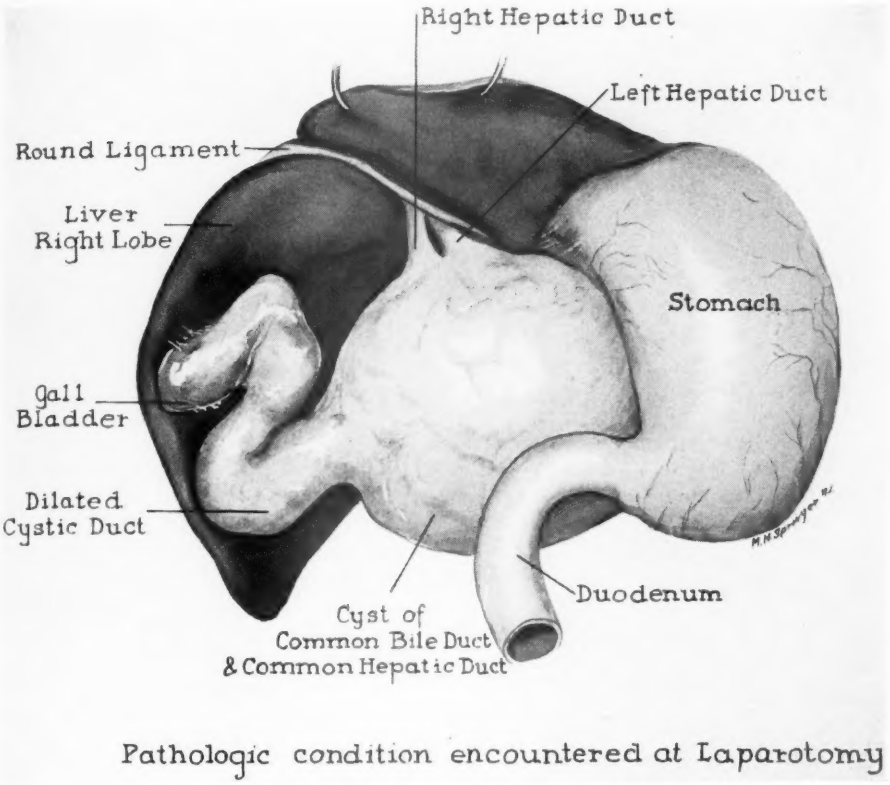


FIG. 6.—Relational anatomy of choledochus cyst.

the transverse colon caudally, and the gallbladder laterally. The peritoneal covering was incised and stripped free, thus liberating the mass at all but three points, namely, the duodenum, gallbladder, and under margin of the liver. The cyst was aspirated and found to contain bile, a specimen of which was sent to the laboratory for study. The gallbladder was slightly distended with inspissated bile, and the cystic duct which opened into the lateral portion of the main mass was greatly enlarged. At the upper surface two dilated separate hepatic ducts opened into the mass. The mass itself opened into the duodenum at the ampulla of Vater through a markedly stenosed lower portion of the common bile duct, the lumen of which was about 1.5 mm. The cystic mass with gallbladder attached was removed (Fig. 7). The left hepatic duct was then anastomosed to the first portion of the duodenum (Fig. 8). The details of this procedure are shown in Figure 9. A No. 18 F. catheter was placed in the left hepatic duct for a distance of about one inch and anchored in place with black silk sutures. A small gastrotomy was then performed near the pylorus and a hemostat introduced. An incision was made

in the first portion of the duodenum over the tip of the hemostat equal in length to the diameter of the hepatic duct. The catheter was then pulled through the incision by the hemostat and the hepatic duct was sutured to the duodenum with black silk sutures. With further traction on the catheter by the hemostat the hepatic duct was pulled into the duodenum for about one half inch, and the serosa of the duodenum was sutured to the duct. This procedure was repeated in anastomosing the right hepatic duct to the first portion of the duodenum (Fig. 10). The catheters passed through the pylorus and lay free in the stomach. The gastrotomy was then closed with black silk sutures. Figure 11 is a plain roentgenogram of the abdomen taken postoperatively showing the catheters in place. The peritoneum and the remainder of the wound were then closed in layers around drains. Four heavy black silk stay sutures were introduced.

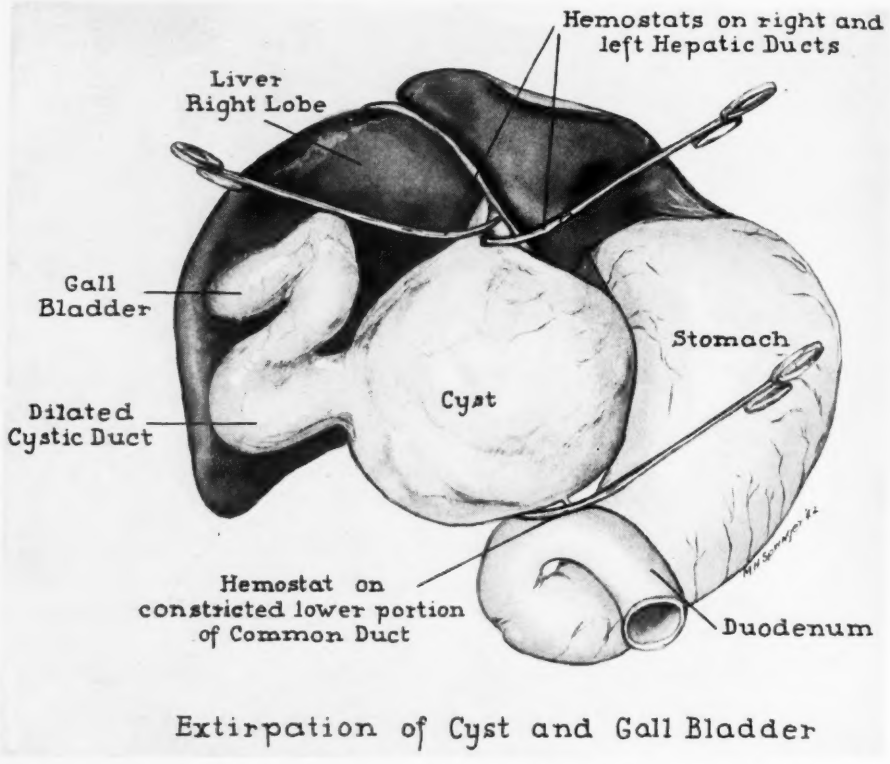


FIG. 7.—Extirpation of choledochus cyst and gallbladder, showing clamps on bile ducts before division.

After one hour and fifteen minutes the anesthesia was supplemented by cyclopropane and ether inhalation by the closed Heidbrink method. The total operating time was two hours and fifteen minutes. The patient left the operating room in good condition. Figure 12 shows the specimen removed at operation.

Pathologic Examination.—Gross: Specimen A consists of a large cyst, 15 cm. in diameter. The wall is greenish-yellow, inelastic and fibrous. It contains serosanguineous fluid. There are no papillae.

Specimen B consists of a gallbladder, measuring 8 x 5 cm., to which the cystic duct, 5 cm. in length, is attached. The gallbladder is thickened and fibrosed and the mucosa is necrotic. It contains greenish, purulent liquid, and no calculi.

The cystic duct is dilated to one centimeter in diameter in certain sections. It is tortuous and its wall is thick and fibrotic. No section of the duct was taken as the specimen is for the museum.

Histologic Examination: Sections of the gallbladder wall are lined with normal columnar epithelium. Many of the cells contain droplets of secretion. The submucosa is moderately edematous, contains many blood vessels and is infiltrated with plasma and round cells. The muscle coats also contain some degree of inflammatory infiltration.

Sections of the common duct (Fig. 13) show a denudation of their mucosa. The mucosal surface is covered with a few plasma cells and lymphocytes. The wall is markedly thickened and composed of dense fibrous connective tissue. There is no evidence of malignancy in any of the sections. *Pathologic Diagnosis:* Marked dilatation of the common duct, cystic duct, and gallbladder; chronic cholangitis; chronic cholecystitis.

Examination of bile removed from the cyst: Material contains a moderate amount of amylase.

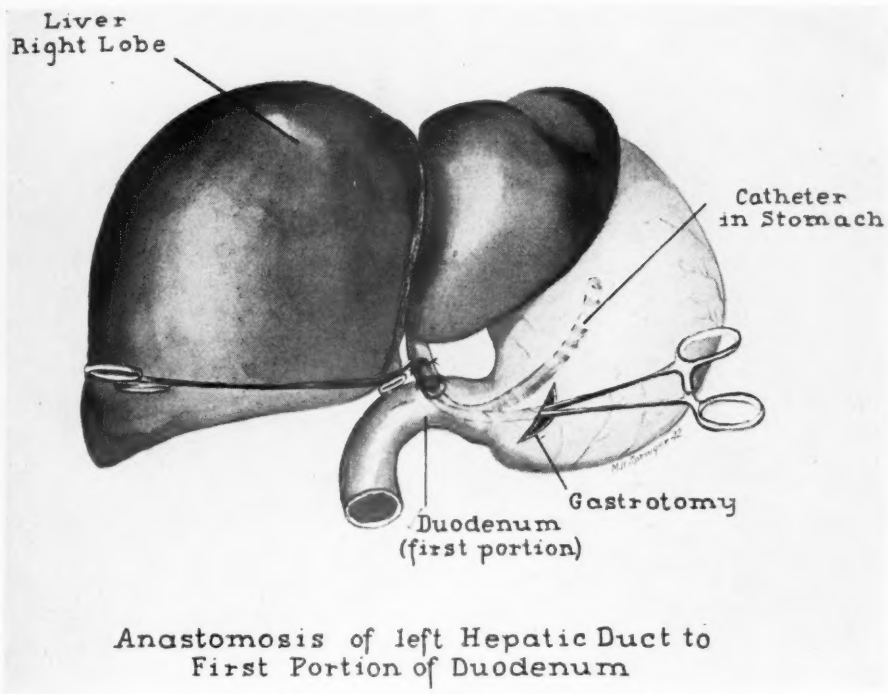


FIG. 8.—Left hepaticoduodenostomy aided by gastrotomy.

Postoperative Course: Aside from the fact that there was rather profuse drainage of bile for a time through the incision, the convalescence of the patient was uneventful. The patient required considerable vitamin K, and in spite of administration several times daily intravenously, the prothrombin level was 25% on the third day postoperatively, 48% on the seventh day, and 52% on the twenty-fifth day. The blood count postoperatively revealed a hemoglobin of 55%; red cells, 2,660,000; and white cells 8,900; color index 1.01. Two blood transfusions of 500 cc. each were given the first week postoperatively. The blood count subsequently rose and just before discharge from the hospital the hemoglobin was 77%; red blood cells, 3,200,000; white blood cells 9,300; color index 1.09.

Routine urinalyses were normal except for an occasional trace of albumin. On the twelfth day the urine was positive for bile pigment and positive for urobilinogen in dilution of 1–40. The color of the stools was normal.

On the eleventh day, liver function studies revealed all dye removed (bromsulfalein

test); positive direct van den Bergh reaction; serum bilirubin, 3.4 mg.; and serum cholesterol was 145 mg.

On the seventeenth day, liver function studies revealed 20% dye retained; positive direct van den Bergh reaction; and serum bilirubin 1.6 mg.

A roentgenogram of the abdomen on the eighteenth day revealed two pieces of rubber tube present in the upper left abdomen, the upper ends being on a level with the twelfth thoracic vertebra and the lower ends extending down to the level of the fourth lumbar vertebra. This study was repeated on the thirty-third day and showed that the two pieces of rubber tube mentioned previously were still present in the upper left abdomen and coincided with the gas shadow of the stomach and were probably within the stomach. They occupied the same position as in the last examination.

On September 6, 1941 (37th day postoperatively), the patient was discharged as improved. Jaundice had disappeared, the operative site was well healed, and the patient asymptomatic. He was instructed to return in six weeks for further study and removal of the tubes.

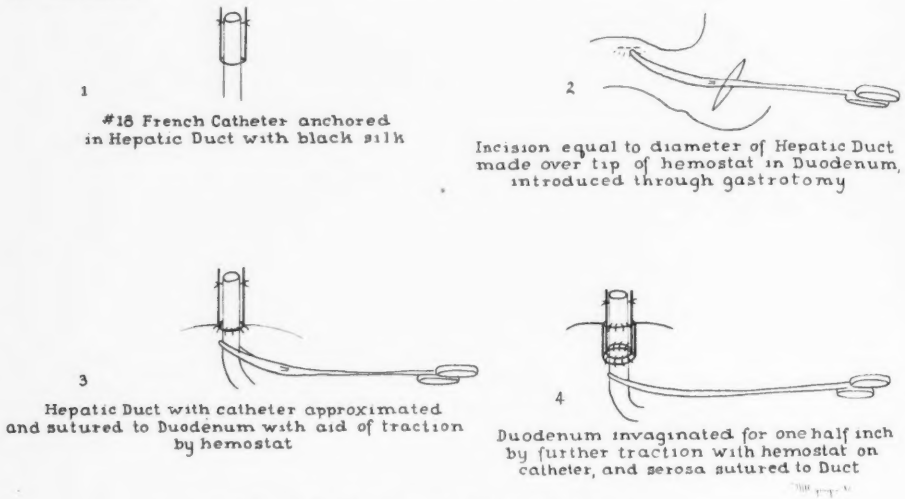


FIG. 9.—Steps employed in hepaticoduodenostomy.

Second Hospital Admission.—The patient was readmitted October 20, 1941. He stated that he had felt well since his discharge from the hospital. The presence of the tubes in his stomach did not cause him distress of any kind. His appetite was good, and he had gained three pounds weight. His weight on this admission was 146 pounds. His bowels were regular and the stools were normal in consistency and color. The remainder of the systemic review was normal.

Physical Examination.—Except for the healed scar of the previous operation in the right upper abdomen, the physical examination was normal.

Laboratory Data.—Blood count revealed hemoglobin 84%; red blood cells, 4,100,000; white blood cells, 8,100; and color index, 0.98. Urinalyses were normal. Liver function studies revealed 10% dye retained (bromsulfalein test); positive direct van den Bergh reaction; and serum bilirubin 1.3 mg.

Reexamination of the upper abdomen roentgenologically again showed the presence of two tubes which overlay the gas shadow of the stomach and were apparently within this organ. The ends of both tubes were visible and apparently both ends also lay within the stomach.

Subsequent Treatment.—On November 3, 1941, a double-plane fluoroscopically assisted gastroscopy was undertaken for removal of the tubes. Two rubber drainage tubes, each about eight inches in length were localized in the stomach along the greater curvature and in the cardia. Each of them was grasped at the distal end with

forward-grasping forceps and removed. It was necessary to reinsert the gastroscope following the removal of the foreign body. A 9 mm. x 53 cm. gastroscope was used.

He was discharged as well November 4, 1941. His weight at that time was 149 pounds. Because of the interest of his case, he was instructed to return in several months for further study.

On June 6, 1942, a plain roentgenogram of the abdomen was negative.

The patient was seen December 7, 1942. He stated that he weighed 155 pounds, had an excellent appetite, and could eat any food without distress. He reported absence of jaundice, and said that his stools and urine were of normal color. His work as a planer in a factory did not cause him to fatigue easily.

For this study 175 cases of congenital cystic dilatation of the common bile duct have been collected from the entire literature. Of this number 82 were taken from the review by Zinninger and Cash[148] (1932). The

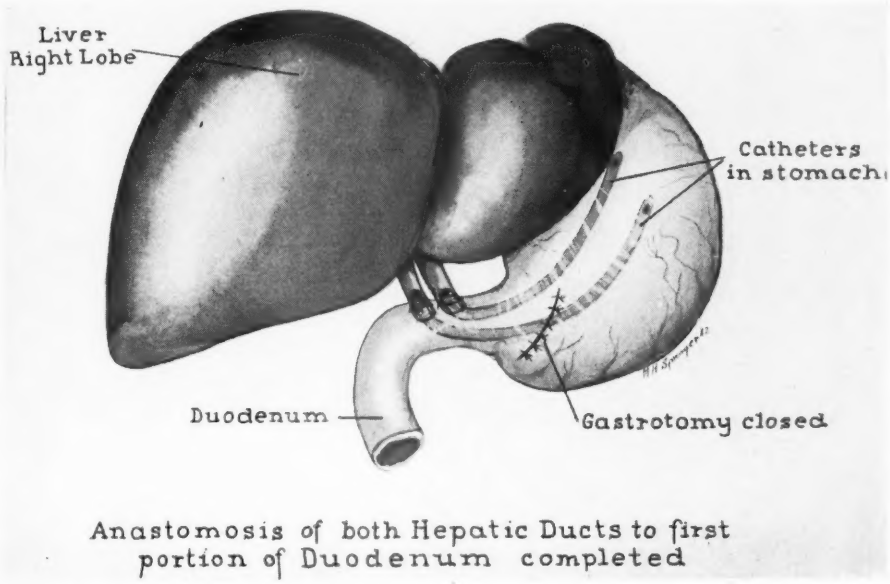


FIG. 10.—Right and left hepaticoduodenostomy completed over retained catheters.

case of Reel and Burrell,[96] included by these authors, is not included in the present review, since much doubt is cast upon its authenticity by Poate and Wade[95] (1941) who believe it should be excluded. From the review by the latter authors 23 added cases are taken. From the review of Japanese literature by Yotuyanagi[144] (1936) 42 cases are included. Of this series 6 were previously included by Zinninger and Cash,[148] and Poate and Wade,[95] so that 36 additional ones appear in the present review. Thirteen of his collected cases are not included. Those of Tanaka,[144] Kawaisi[144] (2nd and 3rd cases), Watanabe,[144] Isimaru[144] (3 cases), and Sioda[144] are excluded because of insufficient or inconclusive data. Those of Terada and Yagi,[121, 144] Kuriyama,[144] Isoda and Kameda,[144] Akamine,[144] and Kawaisi[144] (1st case) are excluded because they seem to represent cases of congenital atresia of the common duct. Two additional cases by Japanese authors and not reported by Yotuyanagi[144] (Murata[86] and Fujihara[50]) are

included, making a total of 44 Japanese cases. The remaining 34 cases constitute a miscellaneous group in which 14 are taken from reports mentioned in reviews by previous authors, 19 cases not analyzed in previous reviews, and the present case report. The latter 20 cases are analyzed in Table V according to the method of Zinninger and Cash[148] (1932) and extended by Poate and Wade[95] (1941).

The most complete reviews of this subject in the past are those of Waller[127] (1917), McWhorter[81] (1924), Seneque and Tailhefer[108] (1929), Zinninger and Cash[148] (1932), Clark[33] (1932), Gross[54] (1933), Yotuyanagi[144] (Japanese literature (1936), Walton[129] (1939), Poate and Wade[95] (1941), and Bangerter[11] (German, 1941).

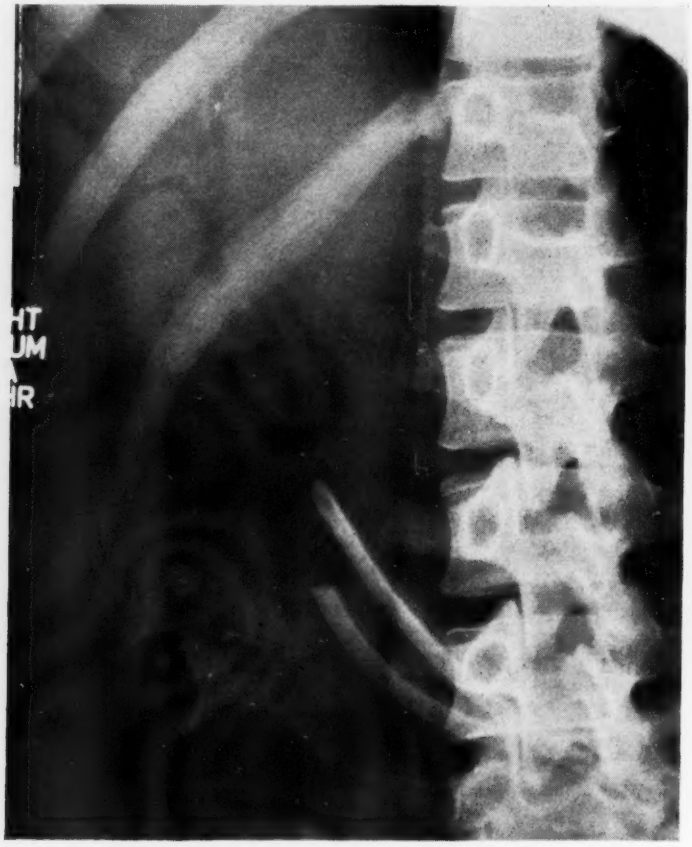


FIG. 11.—Plain roentgenogram of abdomen showing catheters in place.

INCIDENCE

The extreme rarity of this disease is illustrated by the fact that Judd and Greene,[63] in 1926, reported only one case of congenital cystic dilatation of the common bile duct in 17,381 operations on the biliary tract. Furthermore, we have been able to collect only 175 cases. Of these, 44 cases, or 25%, are reported by Japanese authors, so that the disease appears to be relatively more frequent in that country.

In Chart 1 the distribution according to sex and age-group is shown. Females accounted for 77% of the cases. The disease occurs predominantly in children and young adults, since 76% of the patients were below 25 years of age.

ETIOLOGY

Many theories have been advanced to explain the etiology of this unusual lesion, and the following represent the views set forth by the various authors:

1. The cystic dilatation is of "congenital origin" and due to malformation of the choledochus (Giezendanner,[51] Heiliger,[57] Krabbel,[70] Schürholz,[105] and Seeliger[107]).

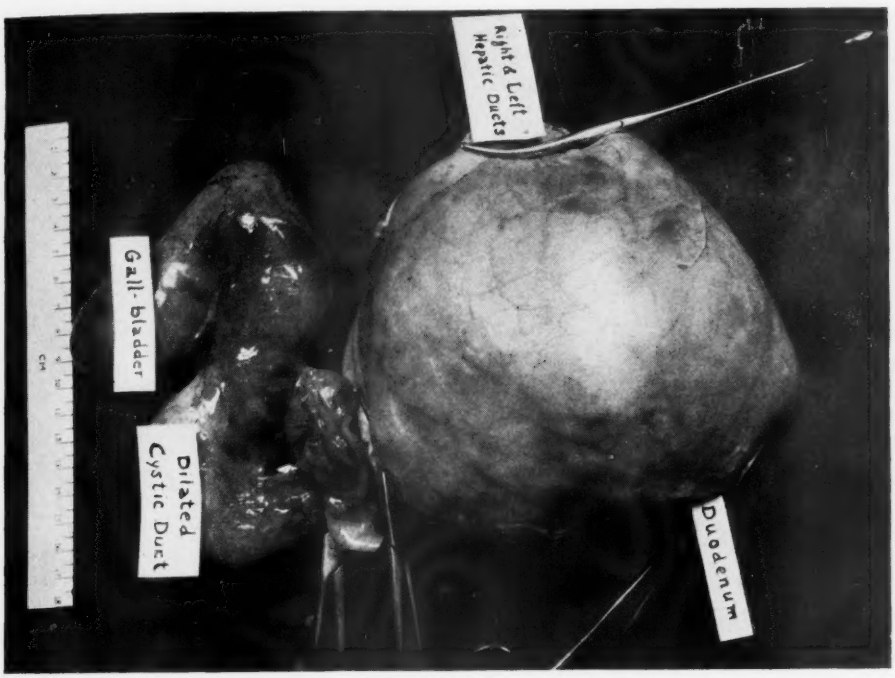


FIG. 12.—Cyst of common bile and common hepatic ducts, with dilated cystic duct and gall bladder.

2. The cyst is caused by an abnormal course of the common duct or angular insertion into the duodenum, congenital in origin, which causes kinking and obstruction (Rostowzew,[99] Arnolds,[4] Konitsky,[68] Ebner,[40] Russell,[101] and Schloessmann[104]).

3. The cyst is the result of congenital narrowing of the intraduodenal portion of the choledochus, or stenosis due to infection (Edgeworth[41] and Ladd[72]).

4. The cyst is caused by a congenital weakness of the wall of the duct (Dreesman,[38] Lavenson,[74] Mayesima,[79] Neugebauer,[88] Kolb,[67] Weiss,[133] Bolle,[22] Russell,[101] and Heiliger[57]). This condition may be similar to congenital idiopathic hydronephrosis in which there may be no demonstrable obstruction (Bohmanson[21]).

5. There is a cystic dilatation on a congenital basis, but with one or more

modifying factors as infection, spasm, kink, valve formation, or stenosis which cause further development of the condition (Zipf[149]).

6. Achalazia may be present at the sphincter of Oddi, causing obstruction by neuromuscular incoordination (Rolleston[97]). Weber[131] agrees with this concept, and believes the condition is explainable on the basis of "autonomic neurodysplasia." The similarity in etiology to megalocolon, megaloesophagus, and megaloureter is suggested, and Weber states that the term "megalocholedochus" is applicable.

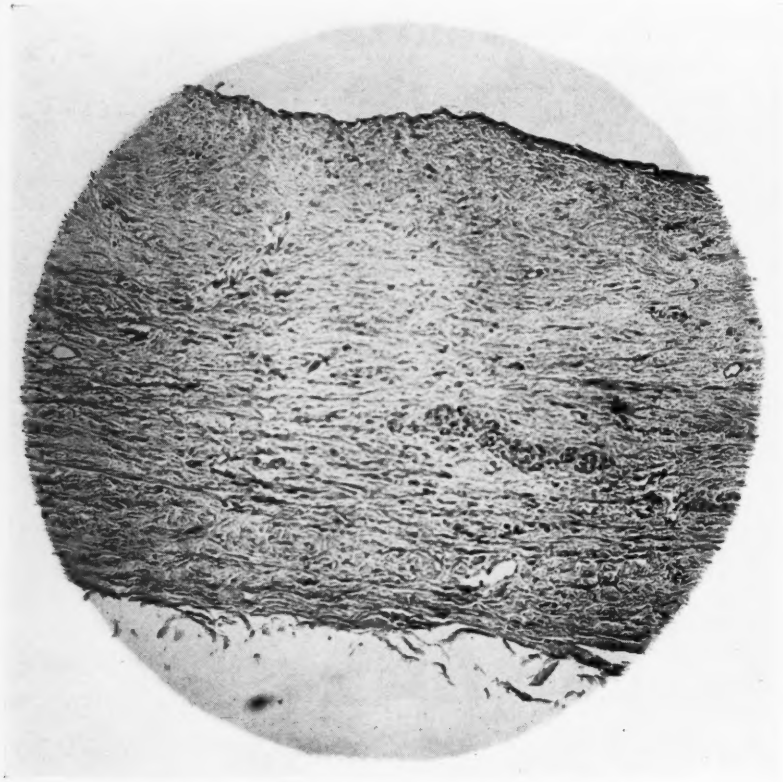


FIG. 13.—Photomicrograph of wall of cyst. (x 100)

7. The cystic dilatation may be caused by the presence of a valve-like fold of tissue over the mouth of the intraduodenal portion of the choledochus (Bakes,[10] Sternberg,[113] Clairmont,[32] Neugebauer,[88] and Rostowzew[99]).

8. Rests of pancreatic tissue in the wall of the choledochus weaken it and permit local dilatation. An analogy is drawn to diverticula of the duodenum in association with aberrant pancreatic tissue (Budde[25]).

9. The cyst may spring from an abortive diverticulum from the choledochus similar to the diverticulum which gives rise to the ventral pancreas (Winternitz[138]).

10. Choledochus cysts may spring from accessory gallbladder formations which are evaginations from the common duct (Budde[25] and Erdely[43]).

11. The cyst may represent a complication of pregnancy as by pressure

from the enlarging uterus (Goldammer[53]), or a kink in the common duct due to the release of intra-abdominal pressure following delivery (McWhorter[81]).

12 The cyst may follow abdominal trauma (Kremer[71]).

13. The cyst may be caused by pressure on the common duct by enlarged tuberculous mesenteric nodes (Douglas[37]).

14. The dilatation may be caused by narrowing of the duct below by persistence of the fetal physiologic epithelial occlusion (Böhm).[20]

15. The etiology lies in the inequality of proliferation of the epithelial cells at the stage when the primitive choledochus is still solid, perhaps at the stage of physiologic epithelial occlusion. If there is a more energetic proliferation of the cells of the upper segment during the stage of physiologic

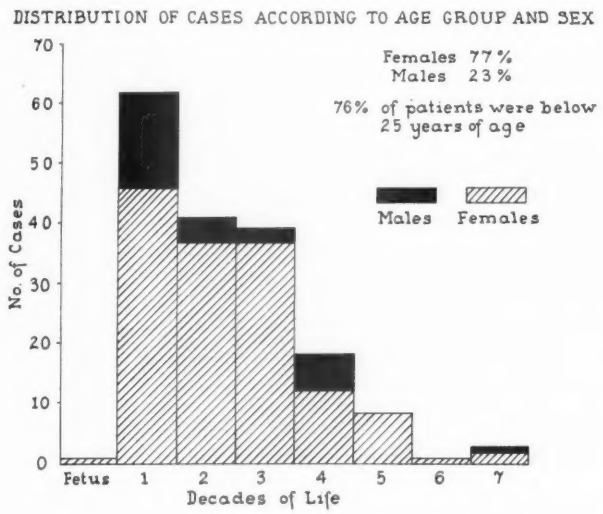


CHART 1.—Distribution of cases according to age group and sex.

epithelial occlusion and less energetic proliferation of the lower segment during the same stage, then, when recanalization occurs later, the upper part of the duct will be abnormally wide and the lower part relatively narrow (Yotuyanagi,[144] 1936).

In consideration of the various theories, it seems most certain that a congenital maldevelopment forms the basis of the abnormality. The theory which appears the most plausible and seems to explain the greatest number of cases is that of Yotuyanagi[144] (1936). His theory is simple, based on sound embryologic principles, and, furthermore, offers a common etiologic basis for the other congenital lesion of the biliary tract with which this lesion is often confused, namely, congenital atresia.

PATHOLOGY

Cystic enlargement of the common bile duct is referred to in the literature as choledochus cyst, cystic dilatation, choledochocoele, diverticulum of the duct, and megalocholedochus. It is also referred to as congenital or idiopathic.

The characteristic pathologic change is a cystic dilatation of that portion of the common bile duct lying above the duodenum. The intraduodenal portion and lower two centimeters of the common bile duct are not involved in the dilatation as a rule. The lesion involves the whole circumference, and is thus not a diverticulum. It is not a cyst in the strict sense of the word, but may be termed "cystic" as it has a relatively thin wall and is full of bile. As a rule, only the common bile duct is involved in the cystic dilatation, so that at the upper pole there are two openings—the common hepatic duct and the cystic duct. In some cases, however, as the case being reported, Cases 1, 2, 3, and 6 of Walton,[129] and in the case of Fowler,[48] the common hepatic duct shares in the formation of the cyst, thus accounting for three openings at the upper pole—the two hepatic ducts, and the cystic duct. At the lower pole the cystic dilatation communicates with the duodenum by means of the lower uninvolved part of the choledochus.

The pathologic findings in congenital cystic dilatation differ from those found in obstruction of the common bile duct by internal or external pressure. The cyst is spherical but always eccentric and thus differs from the uniform dilatation seen above an obstruction. Furthermore, in the congenital type the enlargement is usually localized, whereas in the common types due to stone, tumor, traumatic stricture, or pancreatitis the extrahepatic and intrahepatic ducts and often the gallbladder enlarged as a whole above the site of obstruction.

The cyst may vary in size from a walnut to a mass larger than a full term pregnancy. The amount of fluid may vary from 30 cc. to several liters. In Yotuyanagi's[144] second case the cyst contained 5,200 cc. and Fukada's[144] first case contained 5,000 cc. In the case of Reel and Burrell,[96] disputed by Poate and Wade[95] (1941) as not being an authentic case, 8,000 cc. are reported. There is little relationship between the duration of symptoms and the size of the cyst. In Bolle's case the cyst was "larger than a man's head," and symptoms had been present for only two weeks. In Krabbel's[70] case the cyst was "the size of a fist," and symptoms had been present for eight years.

The cyst wall is usually thickened, seldom thinned out. It may vary from 2 to 7.5 mm. in thickness. The characteristic histologic picture of the common duct is usually lost, so that only a fibrous sack composed of dense connective tissue remains. Sometimes elastic fibers and smooth muscle elements are present, and rarely glands. The lining of the cyst usually lacks epithelium, but cylindrical or cuboidal epithelium may be present. The inner surface may be roughened and coated with heavy irregular deposits of bile pigments. In infected cases there may be purulent exudate on the inner wall and sometimes ulceration. As a rule the cyst wall shows microscopic evidence of extensive inflammatory reaction.

Some authors have described valve-like folds within the cyst. When these thin flaps of tissue are present, they are adjacent to the openings of the ducts. These valves may be several millimeters thick and 1.5 to 2 cm.

in diameter. These flaps when present are found most commonly near the exit of the cyst, but may be adjacent to the hepatic or cystic duct openings.

The cyst contains bile, the character of which no doubt depends on the amount of obstruction present. The bile is apt to be white if obstruction is nearly complete, due to failure of liver function. On the other hand, it may be thick and viscid if obstruction is incomplete and the concentrating power of the gallbladder is retained. In a minority of cases the fluid is clear, thin, and greenish-yellow. The fluid may be sterile in some cases or contain bacteria in others. Schürholz[105] found streptococci and staphylococci as well as gram-negative cocci and rods, Bolle,[22] streptococci, Neugebauer[88] and Walzel,[130] typhoid bacilli, Saint,[102] colon and typhoid bacilli, Zinninger and Cash,[148] paratyphoid bacilli, and Stoney,[114] colon bacilli. In Bangerter's[11] first case the contents were sterile, and in the second case there were gram-positive and gram-negative rods. Stones in the cyst are found only exceptionally (cases of McWhorter,[81] Winterstein,[139] and Sato[144]).

The ducts at the upper pole of the cyst are usually greatly dilated, but this dilatation is cylindrical and does not constitute a part of the cyst. This dilatation rarely extends up to the intrahepatic ducts. In contrast to the dilatation of the ducts at the upper pole is the characteristic constriction of the choledochus at the lower pole of the cyst. Great interest and much speculation has centered around this portion of the choledochus, especially with regard to explanation of etiology of the cyst itself and the obstructive symptoms. Although this portion of the duct is usually narrowed, a great variety of conditions may be present. Angulations and kinks are common. Very large cysts may lie immediately against the duodenum in the region of the ampulla of Vater, leaving only a few millimeters of normal duct to join the cyst to the duodenum. If the cyst is in the upper part of the common bile duct, the lower section may be entirely normal. In some of the reported cases (Erdely,[43] Weiss,[133] Ashby,[6] Arnolds,[4] Rolleston,[97] Giezendanner,[51] and Wyllie[142]) this portion of the duct was obliterated. This obliteration is probably inflammatory in origin, and is sharply to be differentiated from those that are congenital. The age of the patients in these cases precluded the possibility of the obliteration being congenital, and there was a period of months or years in each of these cases during which the patients were free of jaundice. It is probable that congenital stenosis existed with superimposed inflammation and obliteration.

Fowler's[48] case is a notable exception in that the lower part of the duct was greatly dilated. Wheeler[135] (1940) reports a peculiar case of a cherry-sized cyst involving only the intraduodenal part of the common bile duct.

As a rule the constriction of the lower part of the choledochus, when it occurs, does not extend down to the ampulla, and the pancreatic duct is unaffected. In the cases of Seeliger[107] and Mayesima[79] the dilatation occurred so low in the choledochus that the pancreatic duct opened directly into the cyst.

The gallbladder is displaced laterally and is usually of normal size, even

if the cystic duct is dilated. Cases in which the gallbladder is dilated are usually those in older patients in whom the process has been of long duration. In some cases the gallbladder is contracted. Very seldom does it contain stones or gravel. It usually contains clear bile, but in some cases may be empty and chronically inflamed.

The pathologic changes in the liver are not uniformly characteristic and no doubt depend on the degree of obstruction. Frequently the liver is enlarged. Cirrhosis is common, the liver being of a firm consistency and having a fine pebbly surface. The intrahepatic ducts are not dilated as a rule. Microscopically, one finds evidence of cirrhosis with increased periportal connective tissue and proliferation of bile capillaries. Infection is frequent, and the portal areas are markedly infiltrated with leukocytes in these cases. Cholangitis is common and may reach the stage of intraductal suppuration. In advanced cases the presence of lesions characteristic of Banti's syndrome are sometimes found (cirrhosis of the liver and splenomegaly) and may possibly be due to pressure on the portal vein by the cyst and chronic obstruction to flow of bile. In these cases ascites may also be present, and may be due to cirrhosis of the liver or pressure of the cyst on the portal vein. In Walton's[129] first case there was ascites but no true cirrhosis; the portal, splenic, and superior mesenteric veins were thrombosed.

When the cyst is of sufficient size the gastro-intestinal tract is displaced by its presence. The enlarging cyst lies behind and above the duodenum and spreads upwards to the liver, downwards towards the pelvis, and laterally towards the right renal fossa. The stomach is displaced downwards and to the left, the duodenum downwards, medially, and anteriorly, and the hepatic flexure downward and anteriorly.

Two very unusual pathologic findings are recorded in the literature. The first is that present in the case of Swartley and Weeder[117] in which a double common bile duct was present with congenital cystic dilatation confined to the right side. The second is that present in the case of Blocker[18] in which there was traumatic rupture of the cyst following the fall of a 14-year-old boy from a swing across the back of a park seat.

SYMPTOMS AND SIGNS

The salient features of the symptoms and signs of congenital cystic dilatation of the common bile duct constitute a much stressed triad—tumor, jaundice, and pain.

The condition seldom begins suddenly. With careful questioning one can usually trace the onset of symptoms back to early childhood. One may then obtain a history of mild attacks of indigestion with slight upper abdominal discomfort or pain, and mild jaundice which subsided and was given no significance. The longer the interval between these mild attacks, so much the more easily are they forgotten. Until actual obstruction to the bile flow occurs or infection, the condition remains symptomless, and the only sign present is a cystic tumor in the right upper quadrant of the abdomen. In

many cases a typical gallstone history is obtained, with attacks of colicky pain in the right upper abdomen, radiating around the right costal margin and toward the right shoulder, accompanied by nausea and vomiting. The attacks become more frequent, and jaundice, intermittent at first, may become permanent. Often, however, the condition may proceed unnoticed by the patient. Jaundice may become more prominent and unaccompanied by pain. Finally, however, there may arise a feeling of pressure in the liver area, vague discomfort in the epigastrium, anorexia, weakness, hemorrhagic tendencies, and eventually cachexia.

Usually one or two of the triad of diagnostic symptoms and signs are outstanding. Seldom are all three outstandingly present or entirely missing. Table I gives the percentage of these in the present series.

Tumor was present in 77% of the cases, absent in 21%, and questionable or not reported in 2%. In very early cases the tumor may be too small to be palpated definitely, and give only a sense of increased resistance or the suggestion of a mass below the right costal margin and in the right flank.

TABLE I.
THE DIAGNOSTIC TRIAD

	Diagnostic Triad					
	Tumor		Jaundice		Pain	
	Cases	%	Cases	%	Cases	%
Present	134	77	122	70	103	59
Absent	37	21	49	28	61	35
Questionable or not reported	4	2	4	2	11	6

On the other hand, it usually becomes palpable during an attack of pain and jaundice. It may be so large as to fill almost the entire abdomen. It is smooth, elastic, mobile, and may or may not move with respiration. It may be slightly tender. Occasionally on this larger tumor a second smaller mass may be felt—the gallbladder. The liver may be slightly enlarged, but is often not palpable.

Jaundice was present in 70% of the cases, absent in 28%, and questionable or not reported in 2%. The jaundice may be slight, intermittent, or intense and of long-standing. In many cases the stools are acholic, and biliary pigments are present in the urine, these findings varying with the degree of jaundice. The jaundice is of the obstructive type, produced apparently by pressure upon or kinking of the intraduodenal portion of the common bile duct. Thus, the qualitative van den Bergh reaction is usually the positive direct type, and the serum bilirubin is elevated above normal limits, depending on the degree of obstruction. The bromsulfalein test may show liver functional damage by varying degrees of dye retention, and the prothrombin time is often very low, thus accounting for the tendency to hemorrhage reported in the cases.

Pain was present in 59% of the cases, absent in 35%, and questionable or not reported in 6%. This is probably due to increased pressure within

the biliary tree, possibly increased by the presence of infection. Infection is common and probably secondary to stagnation of bile. It is similar to empyema of the gallbladder with cystic duct obstruction. The pain is usually experienced in the epigastrium or right upper quadrant of the abdomen. It is often slight and of a nagging, dragging nature, due to pressure, or discomfort from the abdominal mass. Frank colicky pain may occur and be accompanied by nausea and vomiting. There is no relation between the size of the cyst and the severity of the pain.

Vomiting, when present, usually accompanies the colicky pain or the local peritonitis in infected cases. According to Morley[84] it may be the consequence of a partial duodenal ileus caused by pressure of the cyst on the root of the mesentery. According to Walton,[129] vomiting of bile may follow a sudden abundant flow of bile into the duodenum from the cyst.

Fever is absent in the majority of cases but may be present, particularly in cases with infection of the cyst contents or accompanying inflammation of the gallbladder. It may be a manifestation of cholangitis or hepatitis. It is seldom high except in unusual instances.

Signs of acute peritoneal irritation or inflammation denote cases with infection of the cyst contents or an accompanying cholecystitis. Cases with such manifestations were those of Zinninger and Cash,[148] Saint,[102] and the second case of Bangerter.[11] The cyst was perforated in Walton's[129] sixth case, and also in the case of Blocker[18] (due to trauma).

At the time the patient is admitted to the hospital the general condition is often poor. The patients usually present themselves with liver damage, anemia, and occasionally ascites due to pressure on the portal vein or cirrhosis of the liver. Albumin and casts may be present in the urine.

Roentgenologic studies may be of great aid in many instances. A plain film of the abdomen usually shows a faint shadow of the cyst, which in unusual cases of calcification of the wall becomes distinctly visible. Cholecystogram rarely shows the gallbladder, since in most cases this organ will not concentrate the dye sufficiently. The case of Wright[141] (1935) is the first recorded one in which the gallbladder definitely was visible, showing a peculiar pressure deformity produced by the cyst. However, some of the dye may accumulate in the cyst itself and show an intensification of the shadow shown in the plain film. Gastro-intestinal studies, with barium by mouth and by enema, aid in outlining the positions of neighboring parts of the gastro-intestinal tract. The stomach is displaced to the left and downwards, and may show evidence of external pressure along the lesser curvature. The duodenum is usually displaced medially and anteriorly. It may be somewhat dilated, representing duodenal ileus due to pressure of the cyst on the root of the mesentery. The coils of jejunum may be displaced to the left and downward. The hepatic flexure and transverse colon are displaced downward. The intravenous urogram may be normal, show a tendency to retention of the contrast material in the upper part of the right ureter (as in the present case report), or a slight to moderate degree of hydrone-

phrosis of the right kidney with dilatation of the ureter down to the level of the maximum diameter of the cyst (Wright,[141] 1935).

Peritoneoscopy may be undertaken as a diagnostic aid, as shown in the present case report. In experienced hands this procedure is of value and is to be recommended in cases in which the diagnosis is obscure. The danger of puncture or rupture of the cyst on introduction of the instrument should be kept in mind.

DIAGNOSIS

The triad of symptoms and signs—tumor, jaundice, and pain in a child or young adult, especially a female, should make the diagnosis of congenital cystic dilatation of the common bile duct probable. Blood chemistry and urine studies, liver function tests, prothrombin time determinations, roentgenologic studies, and peritoneoscopy should strengthen the diagnosis. It appears that failure of diagnosis is due to the rarity of the condition, and thus the possibility of congenital cystic dilatation of the common bile duct is usually not considered as a cause of the patient's complaints.

TABLE II.

DIAGNOSES MADE IN 175 CASES

	Preoperative Diagnosis	No. of Cases	Percentage
1	No diagnosis or none stated	59	34
2	Miscellaneous incorrect diagnoses	35	20
3	Choledochus Cyst — First diagnostic impression	15	8.6 } 12.6
	Choledochus Cyst — Mentioned as possible or suspected	7	4.0 }
4	Echinococcus Cyst	17	9.7
5	Cholecystitis or Cholelithiasis	15	8.6
6	Pancreatic Cyst or Tumor	12	6.9
7	Stone in Common Duct	6	3.4
8	Retroperitoneal Cyst Tumor, or Sarcoma	6	3.4
9	Hydrops or enlarged Gall Bladder	4	2.3
10	Cyst of the Liver	3	1.7
11	Obstructive Jaundice	3	1.7

DIFFERENTIAL DIAGNOSIS

In the present series of 175 cases the diagnosis was correctly made 15 times, or in 8.6%; it was mentioned as a diagnostic possibility 7 times, or in 4% of the cases. In most cases either no diagnosis was made or several incorrect diagnostic possibilities were mentioned. In some of the cases the diagnosis was not made even at operation, and in these the result was usually fatal.

Table II shows the diagnoses most commonly made. Among the miscellaneous incorrect diagnoses not mentioned in the table were congestion of the liver, cirrhosis of the liver, splenic anemia, tumor of the liver, hepatic carcinoma or sarcoma, liver abscess, biliary obstruction, congenital abnormality or obliteration of the biliary tract, empyema of the gallbladder, malignancy of the gallbladder, gastric carcinoma, intestinal occlusion, intussusception,

mesenteric cyst, cyst of the kidney, Wilm's tumor of the kidney, hypernephroma, hydronephrosis, ovarian cyst, ruptured viscus, perforated ulcer, abdominal neoplasm, retroduodenal tumor, generalized tuberculosis, and congenital heart disease.

Echinococcus cyst of the liver presents symptoms and signs similar to those of choledochus cyst. This disease may be differentiated in some cases by experienced physicians by eliciting the so-called "hydatid thrill." There may be eosinophilia and the complement fixation test may be positive. The most satisfactory test is the intradermal Casoni reaction, which is both rapid and specific.

Congenital cysts of the liver are usually small and multiple, often associated with cystic kidneys, and as a rule manifest their presence in adult life.

Pancreatic cysts at the head of the gland may produce pressure on the common bile duct and render differential diagnosis difficult or impossible. A pancreatic cyst usually remains the same size or may progressively increase, whereas a choledochus cyst may enlarge or decrease in size from time to time. Pancreatic cysts may also be found more to the left side than that found in choledochus cyst.

Retroperitoneal neoplasms are usually characterized by rapid progression toward a fatal issue. In cases of jaundice with pain slight or absent, one must rule out carcinoma of the ampulla of Vater or the head of the pancreas. Intermittency or relatively long duration of symptoms should eliminate malignancy as a strong diagnostic possibility.

Congenital atresia of the bile ducts occurs only in infants. Jaundice is progressive from birth, and death occurs within four to six months. Age is thus the decisive factor in differential diagnosis.

If pain and jaundice are prominent, the possibility of biliary stones must be considered. However, one must remember that gallstones are uncommon in childhood, and that choledochus cyst occurs predominately in patients under 25 years of age. Roentgenologic studies will aid in indicating the presence or absence of stones.

TREATMENT AND RESULTS

The various types of treatment used and the respective end-results are summarized in Table III.

I. With absence of surgical treatment the patients eventually die, and in all previous reviews the mortality has been 100%. Death is due to biliary cirrhosis, cholangitis, hemorrhage, or rupture of the cyst. In this series the mortality is quoted as 95%, since one patient (case of Wright,[141] 1935) was discharged as unimproved following diagnosis by roentgenology. The only complaint was a mass in the right upper abdominal area, and further information is not available as to her further progress. She was advised to refrain from strenuous exercise. Since in some cases the condition was present for years without causing marked impairment of health, there is no reason to believe that this patient will not eventually fall prey

to obstruction and infection. It is obvious that surgical intervention offers the only hope of cure.

II. Aspiration has no benefit whatever and may lead to peritonitis. The mortality is 100%.

III. Drainage of the cyst, with or without cholecystectomy, is unsatisfactory treatment and carries a high mortality (83% in this series). In Kiselev's[65] case the fistula closed after one and one-half years. McConnell[80] reports a case in which the fistula was closed for two months, reopened for eight months, and then closed again. The fistula in Clairmont's[32] case remained open, and the patient died three years later of tuberculosis. In Wheeler's[135] case of cyst of the intraduodenal portion of the common bile duct, the cyst was drained internally by opening the duodenum and incising the cyst. The cases of Sumpter,[115], Matida,[144] and Bangerter[11] (second case) also recovered.

IV. Drainage followed by secondary anastomosis of the biliary system and gastro-intestinal tract was performed in 24 cases, with a mortality of 29%. In one case, in which recovery ensued, the cyst was excised at the first operation. The disadvantages of this two-stage procedure are the

TABLE III.

TYPES OF TREATMENT IN CASES REPORTED IN THE LITERATURE

Type of Treatment	Cases	Recoveries	Deaths	Mortality
I *No surgical treatment*	22	1[a]	21	95%
II *Aspiration*	5	0	5	100%
III *Drainage ± cholecystectomy*	40	7	33	83%
IV *Drainage followed by secondary anast. of biliary system & GI tract*	24	17	7	29%
A. *Drainage followed by secondary anast. of cyst to G.I. tract*	23	16	7	30%
B. *Cyst excised with drainage; secondary anast. to intestine*	1	1	0	0%
V *Primary anastomosis of biliary system and G.I. tract*	60	44	16	27%
A. *Cyst excised with primary anastomosis of ends of duct*	1	1	0	0%
B. *Cyst excised with primary anastomosis of choledochus, common hepatic duct or both hepatic ducts to G.I. tract*	7	4	3	43%
C. *Primary anastomosis of cyst and G.I. tract ± cholecystectomy*	47	34	13	28%
D. *Cholecystenterostomy*	4	4	0	0%
E. *Primary anastomosis of cyst to stomach, gastro-enterostomy and entero-enterostomy*	1	1	0	0%
VI *Miscellaneous*	24	3	20	83%
A. *Resection of cyst wall*	2	1	1	50%
B. *Excision or attempted excision of cyst ± drainage*	10	1[b]	9	90%
C. *Cyst excised; secondary cholecysto-enterostomy*	1	0	1	100%
D. *Insertion of elastic drainage tube between cyst and duodenum*	1	0	1	100%
E. *Fixation of cyst on abdominal wall ± subsequent aspiratioin ± gastro-enterostomy*	4	0	4	100%
F. *Laparotomy & aspiration, secondary cholecystduodenostomy subsequent aspiration & drainage of cyst, subsequent laparotomy & drainage*	1	1	0	0%
G. *Nature of operation not known*	5[c]	0	4	80-100%
(all types) TOTAL	175	72	102	58%
(surgical operative) TOTAL	148	71	76	51%

a-This patient was discharged as unimproved following diagnosis by x-ray. (Wright) 1935 The only complaint was a mass in the right upper abdominal area. No information is available as to her further progress
b-Case of cyst of double common bile duct with cyst of right duct. Cyst was excised.
c-The outcome of one case was not known.

formation of an external biliary fistula, hemorrhagic tendency due to low prothrombin values, and the extra burden of two operations. In cases in which the operative risk is very great, however, as by marked debilitation or presence of infection, drainage for a short time followed by choledocho-duodenostomy as a two-stage procedure may be advisable. In order to prevent hemorrhage during the period of drainage, the patient should receive vitamin K and bile salts in adequate quantities by mouth, and the prothrombin time should be kept within normal limits.

V. Primary anastomosis of the biliary system and gastro-intestinal tract is the treatment of choice and is accompanied by the lowest mortality. In the present series the mortality with this procedure was 27% in 60 cases. In most cases the treatment advocated by Bakes,[10] in 1907, was carried out, namely primary anastomosis of the cyst itself to the duodenum. The remaining dilated duct, however, may harbor regurgitated food and infection, and may lead to severe ascending cholangitis (case of Fowler[18]). Extirpation of the cyst with primary anastomosis to the duodenum is doubtlessly the most physiologic procedure, for it creates a new anatomic union between the liver and duodenum and removes the cyst which harbors infection. This method, however, carries a higher mortality and is advocated therefore only in cases in which the patient is a good operative risk, the cyst large, and preferably in the absence of infection. In cases in which there is free communication between the gallbladder and the cyst, anastomosis of this organ to the small intestine has given excellent results. This was carried out in four cases, with recovery in each.

VI. A review of the miscellaneous procedures used in treatment reveals a mortality of 83%. In most of these cases the true pathologic condition was not recognized even at operation, and thus a proper rationale for the surgical procedure is lacking.

In cases surgically treated the most common causes of death were shock, hemorrhage, asthenia and debilitation, cholangitis, and peritonitis.

In short, the preferred surgical treatment is to anastomose the cyst to the duodenum, or better, if possible, to excise the dilated part of the common duct and anastomose the remainder to the duodenum. The former is a simpler procedure, carries a lower mortality, and is best undertaken when the operative risk is great. In all cases the prothrombin time should be carefully checked preoperatively and brought to as near normal level as possible by the use of vitamin K and bile salts.

PROGNOSIS

A review of the early literature would give a very gloomy outlook as to prognosis. Each subsequent review of the literature, however, reveals a higher percentage of recovery following operation. In 1927, Lange[73] reported 53 cases in which only 9 had recovered following operation (mortality of 83%). Tailhefer[108, 118] found 29 recoveries out of a series of 82 cases in 1929 (mortality of 65%) as opposed to 49 recoveries out of a series of 115

cases in 1934 (mortality of 57%). The present series shows 71 recoveries out of a total of 148 operated cases (mortality of 51%). Walton,[129] in 1939, reported four cases of recovery out of his five cases operated upon.

The factors involved in prognosis are the preoperative recognition of the lesion, the condition of the patient at the time of operation, and the type of therapy instituted. Table IV shows the effect of correct preoperative diagnosis on mortality. Thus, in cases in which the diagnosis was correct or suspected the mortality was 36%, whereas in cases in which the diagnosis was incorrect, or none made, the mortality was 62%. Cases seen late are usually poor surgical risks as a result of liver damage, cachexia, and myocardial degeneration due to prolonged jaundice. Early cases give the best prognosis because the patient's condition will usually permit the proper surgical procedure to be carried out in one stage, and postoperative complications are less frequent. The type of surgical treatment is of the greatest importance, and it has been found that primary anastomosis of the biliary system and gastro-intestinal tract carries the lowest mortality and offers the greatest chance of cure.

TABLE IV.

EFFECT OF CORRECT PREOPERATIVE DIAGNOSIS ON MORTALITY

Preoperative Diagnosis	Cases	Recovered	Died	Mortality
Correct or suspected	22	14	8	36%
Incorrect or none made	153[a]	57	95	62%

a-The outcome in one case was not known.

Not a great deal is known about the late results in cases of recovery following operation, since in most cases nothing further is reported concerning the progress of the patient. Cases reported as well after four years are those of McConnell,[80] Berkley,[15] and Gross[54] (one case). The following authors report cases in which the patient remained well for more than five years: Iselin,[61] Hildebrand,[58] McWhorter,[81] Murata,[86] Walton[129] (second case), Wheeler,[135] and Sumpter.[115] The case of McWhorter[81] survived for 13 years and died of uremia with nephrosclerosis, hypertension, and diabetes. The case of Wheeler[135] died 15 years later, at the age of 80, of a medical ailment.

SUMMARY AND CONCLUSIONS

One hundred and seventy-five cases of congenital cystic dilatation of the common bile duct are reviewed. A case report is included.

The disease predominates in females and occurs principally in children and young adults.

A congenital maldevelopment probably forms the basis of the abnormality. The most plausible explanation is that of Yotuyanagi[144] (1936), according to whom the etiology lies in the inequality of proliferation of the epithelial cells at the stage of fetal physiologic epithelial occlusion of the common bile duct.

TABLE V

CASES NOT ANALYZED IN PREVIOUS REVIEWS

No.	Author	Year	Sex	Age	Tumor	Jaundice	Pain	Preop. Diag.	Treatment	Results
156	Arullani	1936	F.	34	−	+	−	−	1. Drainage of choledochus. 2. Secondary choledocho-gastrostomy.	Recovery
157	Bangerter	1941	F.	22	−	+	+	Obstructive jaundice. Hemorrhagic diathesis.	Choledocho-gastrostomy.	Death on 3rd postoperative day.
158	Bangerter	1941	F.	60	−	+	+	Empyema of the gallbladder. Pericholecystic abscess?	Cholecystectomy and T-tube drainage of the cyst.	Recovery.
159	Bodley	1937	F.	2	+	+	−	Retroperitoneal sarcoma.	1. Choledochostomy and cholecystoduodenostomy. 2. 25 days later separation of previous anastomosis and performance of choledochoduodenostomy.	1. Improvement for 10 days, then recurrence of symptoms. 2. Death on 2nd postoperative day.
160	Bull	1939	F.	42	+	−	+	Retroperitoneal cyst.	1. Celiotomy and aspiration. 2. Cholecystduodenostomy 3 wks. later. 3. Aspiration and drainage of cyst 3 weeks later. 4. Celiotomy and further drainage.	Recovery.
161	Carr	1940	F.	4	+	+	+	Probable pancreatic tumor or mesenteric cyst. Possible cystic dilatation of common bile duct.	Choledocho-duodenostomy.	Recovery.
162	Duff	1934	F.	62	+	−	+	Retroperitoneal cyst. Possible cyst of common bile duct.	Choledocho-jejunostomy.	Recovery.
163	Fujihara	1935	F.	13	+	+	−	−	Choledocho-jejunostomy.	Recovery.
164	Kiselev	1932	F.	33	−	+	+	−	Resection of the cyst wall.	Recovery.
165	Kiselev	1932	M.	15	+	+	+	−	External drainage.	Recovery. Fistula closed after 1½ years.
166	Koch	1936	?	1½	+	−	+	Congenital heart disease.	None.	Died of congenital heart disease.
167	Mendillo and Koufman	1942	F.	1	+	−	−	Wilm's tumor of right kidney.	1. Irradiation. 2. Excision and choledochoduodenostomy. 3. Omentopexy for cirrhosis of the liver.	1. Fever, anorexia, wt. loss, diarrhea. 2. Recovery. 3. Recovery. Pt. alive in Feb., 1941.
168	Masson and Rieniets	1931	F.	8	+	−	−	Mesenteric or hepatic cyst.	Excision of cyst and hepatico-gastrostomy.	Death on 2nd postoperative day.

169	Murata	1935	F.	1½	+	−	−	Idiopathic dilatation of the common bile duct.	Choledochoduodenostomy.	Recovery. Well after 5 years.
170	Tailhefer	1934	F.	23	+	?	?	−	Choledochoduodenostomy.	Recovery.
171	Tailhefer	1934	F.	24	+	+	+	Congenital dilatation of the common bile duct.	Choledochoduodenostomy.	Death on 4th postoperative day.
172	Wheeler	1915 and 1940	M.	65	−	+	+	Biliary obstruction.	Drainage (duodenum opened and cyst incised).	Recovery. Died 15 yrs. later of a medical ailment.
173	Winterstein	1932	F.	64	+	+	+	Calculous cholecystitis.	Cholecystectomy and choledochoduodenostomy.	Recovery.
174	Wright	1935	F.	39	+	−	−	Congenital cystic dilatation of common bile duct.	None. Patient to refrain from strenuous exercise.	Patient discharged as unimproved.
175	Authors' case	1942	M.	20	+	+	+	Retroperitoneal tumor, probably choledochus cyst.	Extirpation of cyst and gallbladder. Primary right and left hepaticoduodenostomy.	Recovery.

The characteristic pathologic finding is a large cystic dilatation of the common bile duct. The ducts at the upper pole of the cyst are usually dilated, whereas at the lower pole the intraduodenal portion of the duct is usually constricted or angulated. Frequently the liver is enlarged or may show biliary cirrhosis.

The salient symptoms and signs form a diagnostic triad—tumor, jaundice, and pain. Acholic stools may occur, and bile may be present in the urine. The symptoms may begin early in life and occur intermittently over a period of years. Roentgenologic studies and peritoneoscopy may be of aid in confirming the diagnosis. The diagnosis was made or suspected in only 22 cases (12.6%), but consideration of the condition as a diagnostic possibility should lead to more frequent correct diagnosis.

The procedure of choice in treatment is a primary anastomosis of the biliary and intestinal tracts. Anastomosis of the cyst itself to the duodenum is accompanied by the lowest mortality, but in good-risk patients extirpation of the cyst was primary anastomosis to the duodenum is preferable because the cyst may harbor infection and regurgitated food. The latter procedure was performed, with recovery, in the case herein reported.

The mortality in the entire series was 58%, but in those patients treated by primary anastomosis of the biliary and intestinal tracts the mortality was 27%. The prognosis depends on the preoperative recognition of the lesion, the condition of the patient at the time of operation, and the type of therapy instituted.

BIBLIOGRAPHY

[1] Abe, S.: Three Autopsy Reports. Tr. Soc. path. jap., **25**, 423–429, 1935. (In Japanese).

[2] Adam, L.: Zentralblatt für Chirurgie, **1**, 997, 1923.

[3] Ahrens: Zentralblatt für Chirurgie, **1,** 672, 1920.

[4] Arnolds: Manneskopfgrossen Retentionscyste des Choledochus. Deutsche med. Wchnschr., **32,** 1804, November 1, 1906.

[5] Arullani, C.: La dilatazione idiopatica del coledoco. Boll. Accad. lancis. Roma, **8,** 643–650, 1936.

[6] Ashby, H.: Medical Chronicle, **10,** 28, 1898.

[7] Bachkiroff: Sur la pathogenie des kystes du choledoque. Chir. Arch. Veliaminova, **27,** 63, 1911.

[8] Backer-Grondahl, N.: Idiopathic Cysts of Common Duct. Med. rev. Bergen, **50,** 337–354, August, 1933.

[9] Badile: Archivio italiano di chirurgia, **51,** 424, 1924.

[10] Bakes: Wien. klin. Wchnschr., **20,** 298, 1907.

[11] Bangerter, J.: So-called Idiopathic Cysts. Schweiz. med. Wchnschr., **71,** 677, May 31, 1941; *idem*, ibid: **71,** 733, June 14, 1941.

[12] Beaufays: Z. org. Chir., **57,** 412, 1932.

[13] Bencevic: Z. org. Chir., **43,** 421, 1928.

[14] Bengolea, A. J., and Suarez, C. V.: Idiopathic Cystic Dilatation of Hepatocholedochus; Case. Nord. med. (Norsk mag. f. laegevi densk), **1,** 31–36, January 7, 1939.

[15] Berkley, H. K.: Idiopathic Dilatation of the Common Bile Duct in Childhood. Jour. of Pediatrics, **14,** 79, 1939.

[16] Berman, J. K., and Ballenger, F.: Congenital Cyst of Common Duct. Quart. Bull Indiana Univ. M. Center, **3,** 141–144, October, 1941.

[17] Bistrov, P. B.: Kharkovskiv Med. Jour., **19,** 319, 1915.

[18] Blocker, T. G. and others: Traumatic Rupture of Congenital Cyst of Choledochus. Arch. Surg., **34,** 695, 1937.

[19] Bodley, J. W.: Choledochus Cyst; Case; South, Surgeon, **6,** 126–130, April, 1937.

[20] Böhm, F.: Ztschr. f. ang. Anat. u. s. w. (Berlin), **1,** 105, 1913.

[21] Bohmanson, C.: Cyst of the Common Bile Duct. Acta Chirurgica Scandinavica, **56,** 440, 1924.

[22] Bolle, H.: Ein Fall von idiopathischer Choledochuszyste. Deutsche med. Wchnschr., **48,** 1381, October 13, 1922.

[23] Broca: Bull. et mem. Soc. nat. de chir., **23,** 209, 1897.

[24] Brun and Hartman: Bull. et mem. Soc. nat. de chir., **23,** 207, 1897.

[25] Budde, M.: Über die Pathogenese und das Krankheitsbild der cystischen Gallergangerweiterung (sogennante idiopatische Choledochuscyste). Deutsche Ztschr. f. chir., **157,** 364, 1920.

[26] Bull. P.: Idiopathic Dilatation of Choledochus (Congenital Cyst); Case. Nord. med. (Norsk mag. f. laegevi densk), **1,** 31–36, January 7, 1939.

[27] Burman, G. E. L.: A Case of Simple Obstructive Jaundice with Huge Dilatation of the Common Bile Duct. The Practitioner, Page 473, March, 1912.

[28] Butters: Choledochus-Zysten. Deutsche med. Wchnschr., **36,** 1351, July 14, 1910.

[29] Carr, G. L.: Congenital Cystic Dilatation of Common Bile Duct. M. Woman's J., **47,** 301–303, October, 1940.

[30] Chiray, M., Albot, G., and Jame, R.: Obstructive Jaundice due to Stenosis of Vater's Ampulla and Cystic Dilatation of Bile Duct in a Girl Seven Years Old. Gaz. med. de France (supp. Gastro-enterol.), Pages 3–6, November 15, 1933.

[31] Choyce: Cited by Stoney.[114]

[32] Clairmont: Zystische Dilatation des Ductus Choledochus. Wien klin. Wchnschr., **24,** 1616, 1911.

[33] Clark, J. H.: So-called Idiopathic Choledochus Cyst; Pathogenesis and Pathology; Report of Case with Review of Literature. Internat. Clin. **1,** 78–103, March, 1932.

[34] De Quervain: Spezielle chirurgische Diagnostic, Page 398, 1922.

[35] Derwissien: Zentralblatt für Chirurgie, **54,** 852, April 12, 1927.

36 Di Natale, L.: Cisti del coledoco, Arch. ital. di chir., **23,** 553, May, 1929; abstr. Brit. M. J., **2,** 30, August 24, 1929.

37 Douglas: Monthly J. M. Sc. (London), **14,** 97, 1852; cited by McWhorter,[81] and Dreesman.[38]

38 Dreesman: Beitrag zur Kenntnis der kongenitalen Anomalien der Gallenwege. Deutsche Ztschr. f. Chir., **92,** 401, 1908.

39 Duff, D. D.: Cyst of the Common Bile Duct. Brit. Jour. Surg., **21,** 536, 1933–34.

40 Ebner, A.: Idiopathische Choledochuscyste und Purpura Hemorrhagica Fulminans. Beitr. Z. klin. Chir., **64,** 473, 1909.

41 Edgeworth, F. H.: A Case of Dilatation of the Common Bile Duct Simulating Distention of the Gallbladder. Lancet, **1,** 1180, May 11, 1895.

42 Elischer: Zentralblatt für Chirurgie, **9,** 341, 1923.

43 Erdely: Deutsche Zeitschrift für Chirurgie, **209,** 65, 1928.

44 Ewoyan: Cited by Sebek.[106]

45 Exner, A.: Choledochus Cyst. Wiener klinische Wochenschrift, **24,** 1615, 1911.

46 Feyrter, F.: Über Fehlbildungen der extrahepatelen Gallenwege mit Störung der normalen Hohlraumbildung (Verödung, Enge und cystische Erweiterung) und des normalen Gangverlaufes. Virchows Arch. f. path. Anat., **271,** 20, 1929.

47 Flechtenmacher, C.: Operativ entfernte Choledochuszyste im Ligamentum hepatoduodenale. Wien. klin. Wchnschr., **32,** 365, 1919.

48 Fowler, R. S.: Choledochus Cyst. Annals of Surgery, **64,** 546, 1916.

49 Frizelle, E. R.: Idiopathic Congenital Dilatation of the Common Bile Duct. Brit. Med. Jour., **1,** 610, 1932.

50 Fujihara: Arch. jap. Chir. (Kyoto), **12,** 1206–1212, 1935; abstr. in Z. org. Chir., **75,** 117–118, 1936.

51 Giezendanner, E.: Beitrag zum Studium der kongenitalen Choledochuszystem, Frankfort. Ztschr. f. Path., **38,** 150, 1929.

52 Gokhman, A. I.: Idiopathic Cyst of Common Bile Duct; Case. Novy khir. Arkhiv., **37,** 486–489, 1936.

53 Goldammer: Beiträge zur Chirurgie der Gallenwege. Beitr. z. klin. Chir., **55,** 214, 1907.

54 Gross, R. E.: Idiopathic Dilatation of the Common Bile Duct in Children. Jour. Ped., **3,** 730, 1933.

55 Hamilton, I.: Congenital Cystic Dilatation of the Common Bile Duct. Med. Jour. of Australia, Page 800, May 27, 1939.

56 Heid: Inaugural Dissertation. Cited by McWhorter.[81]

57 Heiliger: Inaugural Dissertation. Cited by McWhorter.[81]

58 Hildebrand: Geheilte Choledochuszyste. Deutsche med. Wchnschr., **39,** 2224, November 6, 1913.

59 Hill, N. H., and Ramsay, R. A.: Cystic Dilatation of the Common Bile Duct. Brit. M. J., **2,** 991, November 28, 1925.

60 Ipsen, J.: Et tilfaelde af choledochuscyste. Hospitalstid, **45,** 1342, 1913. Cited by Waller.[127]

61 Iselin, H.: Gemeinsame Cyste der Gallen-und Pankreaswege. Arch. f. klin. Chir., **145,** 304, 1927.

62 Jauregui, P.: Cyst of the Common Bile Duct. Rev. de cir., **7,** 536, 1928.

63 Judd, E. S., and Greene, E. L.: Choledochus Cyst. Surg. Gynec. and Obst., **46,** 317, 1928.

64 Keyrovsky: Inaugural Dissertation. Greifswald, 1888 (in Seyffert[109]).

65 Kiselev, N.: Idiopathic Dilatation of the Common Bile Duct. Vestn. Chir., **78/79,** 182–186, 1932.

66 Koch, F.: So-called Choledochus Cyst (Idiopathic Congenital Dilatation). Hygeia **98,** 577–587, September 15, 1936.

67 Kolb: Inaugural Dissertation. Cited by Waller[127] and McWhorter.[81]

68 Konitsky: Inaugural Dissertation; cited by Waller; cited by McWhorter.

69 Kőrte: Die Erkrankungen der Gallenwege. Med. Praxis, Page 117, 1928. Cited by Seneque and Tailhefer.[108]

70 Krabbel, M.: Zum operativen Heilung der idiopathischen Choledochuscyste. Beitr. z. klin. Chir., **130,** 159, 1924.

71 Kremer, J.: Ein Fall einer durch Choledocho-Duodenostomie dauernd geheilten echten Choledochuszyste. Arch. f. klin. Chir., **113,** 99, 1919.

72 Ladd: J. A. M. A., **91,** 1082, 1928.

73 Lange, K.: Beitrag zur idiopathischen Choledochuscyste. Zentralbl. f. Chir., **54,** 2287, 1927.

74 Lavenson, R. S.: Cysts of the Common Bile Duct. Am. J. M. Sc., **137,** 563, 1909.

75 Letulle, M.: Dilatation kystique des voies biliares cholangiectasies congenitales. Presse med., **21,** 97, February 1, 1913.

76 Limito, C.: Idiopathic Cyst of Choledochus; Case. Arch. ital. di anat. e. isto., pat., **6,** 619–627, Sept.–Oct., 1935.

77 Masson, J. C., and Rieniets, J. H.: Adenocarcinoma and Squamous Cell Carcinoma in a Cyst of the Mesosigmoid (Case Report); Choledochus Cyst (Case Report). S. Clin. North America, **11,** 743–751, August, 1931.

78 Matthieu, J.: Congenital Cyst of the Common Bile Duct. ANNALS OF SURGERY, **92,** 159, 1930.

79 Mayesima, J.: Zur Kasuistik der Primären cystischen Erweiterung des Ductus Choledochus. Deutsche Ztschr. f. Chir., **119,** 338, 1912.

80 McConnell, A.: Cyst of the Common Bile Duct. Brit. J. Surg., **7,** 520, April, 1920.

81 McWhorter, G. L.: Congenital Cystic Dilatation of the Common Bile Duct. Arch. Surg., **8,** 604, March, 1924.

82 Melichoff: Dilatation idiopathique du choledoque. Russkaya klinika, **3,** 74, 1925.

83 Mendillo, A. J., and Koufman, W. B.: Choledochus Cyst; Case. Connecticut M. J., **6,** 99–101, February, 1942.

84 Morley, J.: Congenital Cyst of the Common Bile Duct. Brit. Jour. of Surg., **10,** 413, 1923.

85 Morrin: cited by Stoney.

86 Murata: Über einen dauernd geheilten Fall von idiopathischer Choledochuserweiterung. Zbl. f. Chir., **62,** 1269–1271, 1935.

87 Narabayashi: Z. org. Chir., **25,** 401, 1924.

88 Neugebauer, F.: Zur Kenntnis der idiopathischen Choledochuscyste. Beitr. z. klin. Chir., **131,** 448, 1924.

89 Nicolaysen, J.: Zentralbl. f. Chir., **16,** 1269, 1899.

90 Oglobin, A. A.: Idiopathic Cysts of the Bile Ducts. Vestnik khir., **9,** 177, 1927; abstr. Internat. Abstract Surg., Page 324, October, 1928.

91 O'Gorman, F. M., *et al.*: Idiopathic Choledochus Cyst; Report of a Case, New York State Jour. of Med., **31,** 1196, 1931.

92 Olivecrona, H.: Infected Cyst of the Common Bile Duct; Operation; Recovery. Acta chir. Scandinav., **62,** 347, 1927.

93 Oxley, M. G. B.: Congenital Atresia of the Duodenal Opening of the Common Bile Duct in an Infant, Producing a Large Abdominal Tumor. Lancet, **2,** 988, December 8, 1883.

94 Pamperl, R.: Medizinische Klinik, **21,** 1608, 1925.

95 Poate, H. R. G., and Wade: Congenital Cystic Dilatation of the Common Bile Duct. Australian and New Zealand J. Surg., **11,** 32–45, July, 1941.

96 Reel, P. J., and Burrell, N. E.: Cystic Dilatation of the Common Bile Duct. ANNALS OF SURGERY, **75,** 191, 1922.

97 Rolleston, H. D.: Diseases of the Liver, Gallbladder and Bile Ducts. ed. 3, New York, The Macmillan Co., 1905, p. 651.

98 Rosenberg: Archives für klinische Chirurgie, **125,** 1–2, July 31, 1923.

99 Rostowzew, M. J.: Ein Fall von hochgradiger cystischer Erweiterung des Ductus Choledochus. Deutsche med. Wchnschr., **28,** 739, October 9, 1902.

100 Rotgans: cited by McWhorter.

101 Russell, R. H.: Case of Cystic Dilatation of the Common Bile Duct in a Child. ANNALS OF SURGERY, **26,** 692, 1897.

102 Saint, J. H.: Cystic Dilatation of the Common Bile Duct. Brit. Med. Jour., p. 230, 1932.

103 Santrucek, K.: Idiopathic Cyst of Bile Duct. Casop. lek. cesk., **71,** 1185–1186, September 16, 1932.

104 Schloessmann: Beitrag zur Kenntnis der Choledochuscysten. Deutsche Ztschr. f. Chir., **109,** 160, 1911.

105 Schürholz: Ein Fall von sogenannten idiopatischer Choledochuscyste. Arch. f. klin. Chir., **118,** 91, 1921.

106 Sebek: O pricinach ysiniku, pathologii a sympotomologii tak zvane idiopathicke cysty choledochu. Casop. lek. cesk., **66,** 1720, 1927. Cited by Seneque and Tailhefer.

107 Seeliger, S.: Beigrag zur Kenntnis der echten Choledochuscysten. Beitr. z. klin. Chir., **99,** 158, 1916.

108 Seneque, J., and Tailhefer, A.: Les dilatations congenitales due choledoque. J. de chir., **33,** 154, 1929.

109 Seyffert. Cited by McWhorter.

110 Sieber, F.: Large Cyst of the Bile Ducts. Zentralbl. f. Gynäk., **53,** 2848, 1929.

111 Smit: Medicinisk Revue, **32,** 285, 1915.

112 Söderlund: A Case of Stone in the Bile Ducts. Acta chir. Scandinav., **59,** 253, 1925.

113 Sternberg, C.: Cholezystostomie. Wien. klin. Wchnschr., **24,** 1616, 1911.

114 Stoney, R. A.: Cyst of the Common Bile Duct. Irish J. M. Sc., page 764, December, 1929.

115 Sumpter, W. D.: South. Pract., **39,** 169, 1917.

116 Swain, W. P.: A Case of Cholecystenterostomy with the Use of Murphy's Button. Lancet, **1,** 743, March 23, 1895.

117 Swartley, W. B., and Weeder, S. D.: Choledochus Cyst with a Double Common Bile Duct. ANNALS OF SURGERY, **101,** 912, 1935; see further report on the same case by Weeder.

118 Tailhefer, A.: Dilatation Congenitale du canal choledoque. Bull. Soc. nat. Chir. Paris, **60,** 8–11, 1934.

119 Tavernier, M.: Dilatation congenitale du choledoque. Lyon Chir., **31,** 595–597, 1934.

120 Taylor, J.: Cystic Dilatation of the Common Bile Duct: Record of an Example. Brit. J. Surg., **16,** 327, 1928.

121 Terrada and Yagi: Jour. of Ped., Tokyo, **317,** 2078, 1930; reviewed in Am. Jour. Dis. of Child., **42,** 1256, 1931.

122 Ugelli, L.: Sulle dilatazioni congenite del coledoca. Policlinico (sez. chir.), **40,** 343–355, 1933.

123 Upcott, H.: Cystic Dilatation of the Common Bile Duct. Brit. Med. Jour., Page 911, 1932.

124 Vara-Lopez, R., and Meana Negrete, V.: Congenital Cystic Dilatation of Choledochus: Report of Cases. Med. espan., **3,** 471–483, June, 1940.

125 Virgillo, F.: Congenital Cystic Dilatation of Choledochus; Case, Arch. ital. di. chir., **54,** 873–878, 1938.

126 Wagner, A.: Beitrag zur Chirurgie der Gallenwege. Deutsche Ztschr. f. Chir., **145,** 15, 1918.

127 Waller, E.: Idiopathic Choledochus Cyst. ANNALS OF SURGERY, **66,** 446, 1917.

[128] Walters and Snell: Diseases of the Gallbladder and Bile Ducts. Saunders Co., 1940, Pages 236–240.

[129] Walton, J.: Congenital Diverticulum of the Common Bile Duct. Brit. Jour. of Surg., **27,** 295, October, 1939.

[130] Walzel, P.: Zentralblatt für Chirurgie, **48,** 594, 1921.

[131] Weber, F. P.: Cystic Dilatation of the Common Bile Duct. Brit. Jour. of Child Dis., **31,** 27 and 113, 1934.

[132] Weeder, S. D.: Choledochus Cyst with Double Common Duct; Sequelae and Complications. ANNALS OF SURGERY, **110,** 55–59, September, 1939.

[133] Weiss, S.: Ein seltener Fall von cystischen Erweiterung des Ductus Choledochus. Berl. klin. Wchnschr., **46,** 1843, 1909.

[134] Wettwer: Ein Fall von congenitalen Choledochuscyste: Dissertation, Göttinger, 1907; cited by Waller.

[135] Wheeler, W. I. de C.: Dublin Jour. Med. Sci., December, 1915; Unusual Case of Obstruction to Common Duct (Choledochocele?). Brit. J. Surg., **27,** 446–448, January, 1940.

[136] Willis, B. C.: Congenital Cystic Dilatation of the Common Bile Duct. ANNALS OF SURGERY, **87,** 48, 1928.

[137] Wilson, H. V., Jr.: Choledochus Cyst. J. A. M. A., **95,** 399, August 9, 1930.

[138] Winternitz; quoted by Erdely.[43]

[139] Winterstein, O.: Bile Duct Cysts: Diagnosis. Wien, med. Wchnschr., **82,** 638–639, May 21, 1932.

[140] Wright, H. W. S.: Congenital Diverticulum of the Common Bile Duct. Surg., Gynec., and Obst., **39,** 156, 1924.

[141] Wright, A. D.: X-ray Appearances Produced by Congenital Cystic Dilatation of Common Duct. Brit. J. Radiol., **8,** 227–230, April, 1935.

[142] Wyllie, W. G.: Congenital Cyst of the Common Bile Duct. Lancet, **1,** 1342, 1925.

[143] Yamanouchi, K.: Über die cystische Dilatation des Ductus choledochus. Ztschr. f. Jap. Chir. Gesellschr., **22,** 1921; abstr., Zentralorg. f. d. ges. Chir., **16,** 228, 1922. Cited by Neugebauer, Judd and Greene, and Willis.

[144] Yotuyanagi: Contribution of the Etiology and Pathogeny of Idiopathic Cystic Dilatation of the Common Bile Duct. Gann., **30,** 601, October, 1936.

[145] Young: Glasgow Medical Jour, **107,** 346, 1927.

[146] Zaczek: Congenital Cyst of the Common Bile Duct. Polski Prezegl. Chir., **6,** 639, 1927.

[147] Zimmer, H.: Zentralblatt für Chirurgie, **51,** 424, 1924.

[148] Zinninger and Cash: Congenital Cystic Dilatation of the Common Bile Duct. Arch. Surg., **24,** 77–105, January, 1932.

[149] Zipf, K.: Über idiopathische Choledochuscysten. Arch. f. klin. Chir., **122,** 615, 1922.

Note: For references for 42 of the cases taken from the Japanese literature, see the review by Yotuyanagi[144] (1936).

THE INDICATIONS FOR JEJUNAL ALIMENTATION IN THE SURGERY OF PEPTIC ULCER*

RALPH COLP, M.D., AND LEONARD J. DRUCKERMAN, M.D.
NEW YORK, N. Y.

FROM THE SURGICAL SERVICE, MOUNT SINAI HOSPITAL, NEW YORK, N. Y.

THIS REPORT comprises a series of 51 ward patients upon whom jejunostomy was performed for alimentation. This group consisted mainly of those in whom recurrent chronic peptic ulceration had left serious indelible physical imprints. The average age was 48, and the average duration of the disease was almost 12 years. Sixteen patients had experienced one or more attacks of severe gastro-intestinal hemorrhage. Many had been previously operated upon, 14 had been explored for an acute perforation of an ulcer, seven had had a gastro-enterostomy, and four a subtotal gastrectomy. Some of the patients were admitted to the hospital with a definite alkalosis, and others entered during an episode of severe hemorrhage. The accumulated experiences gained from a study of a group such as this, crystallized quite clearly, the indications for jejunostomy for alimentation in the surgery of peptic ulceration.

Jejunostomy was employed as a conservative preliminary procedure in uncontrollable alkalosis and in ulceration too extensive for immediate radical gastric surgery. It was established as a definitive palliative measure in those cases of severe peptic ulceration in which, because of certain physical conditions, any other type of surgery was deemed inadvisable. It was used quite frequently as a complement to gastric operations in a prophylactic effort to avoid the dire consequences of gastric atony, and finally, it was utilized as a supplementary procedure in those postoperative cases in which an intractable gastric ileus, with alkalosis developed.

PRELIMINARY JEJUNOSTOMY

This has been successfully employed by Balfour and Eusterman[6] in those cases of massive gastric ulceration in which radical surgery was impossible at the time of operation. The benefits derived from this simple procedure were so marked that in many instances subtotal gastrectomy could be performed at a later period. Preliminary jejunostomy has been used in this clinic in some complicated cases of pyloric and duodenal stenosis with gastric dilatation. The symptom-complex of this condition was characterized mainly by repeated vomiting or the retention of large amounts of ingested food and gastric secretions. The inability to retain water, food and vitamins, and the loss of the acid secretion of the stomach caused fundamental alterations in the chemistry of the blood, notably an increase in the urea nitrogen, a diminution in the chlorides, and an increase in the carbon dioxide combining

* Read before the New York Surgical Society, December 9, 1942.

power. This pattern is called alkalosis. It has serious implications, for severe alkalosis is incompatible with life. Preliminary jejunostomy was performed for alimentation in six patients, because of a pronounced alkalosis which did not respond to the usual conservative therapy. The latter consisted of the parenteral administration of adequate amounts of saline and glucose which, as a rule, temporarily corrected the hypochloremia, provided that the kidney function was normal. The amount of salt in solution necessary to raise the lowered chloride content of the blood to normal limits may be measured by the formula of Coller, *et al.*[4] The lowered serum proteins frequently found in the starvation incident to long-standing gastric obstruction were often effectively elevated by the administration of blood, blood plasma and amino-acids in sufficient quantities. In addition, the stomach was lavaged twice a day with hot saline solution in an effort to eliminate stagnation and diminish intragastric tension. This procedure often allayed the inflammatory reaction about an ulcer and relieved pylorospasm to such an extent that gastro-intestinal continuity was established and the stomach partially regained its tone. The efficacy of this treatment was gauged, empirically, by a reduction in the amount of gastric retention and, chemically, by a diminution in the degree of alkalosis. However, these conservative measures were found to be futile if persistent vomiting caused a constant loss of ingested food, fluid and gastric secretions. This type of high intestinal obstruction demanded surgical relief. Under these conditions, major gastric surgery was definitely contraindicated and, therefore, a preliminary jejunostomy for alimentation was performed under local anesthesia. Jejunal alimentation usually restored nutrition and hydration, and corrected alkalosis within a short period of time.

Beside the correction of the alkalosis and the subsequent improvement in the physical condition of the patient, a preliminary jejunostomy served other important functions. Jejunal alimentation, by eliminating gastric digestion, permitted the atonic stomach, now partially defunctionalized, to regain its tone and gradually contract. In addition, it allowed the local inflammatory reaction about an extensive penetrating ulcer to subside to such a degree that a subtotal gastrectomy could be performed eventually without undue risk.

The time intervening between the preliminary jejunostomy and subsequent gastric surgery varied. It was dependent upon the clinical improvement of the patient, the diminution in the amount of gastric retention, the amelioration of the alkalosis, and a recession in the extent of the local pathology, as demonstrated roentgenologically. The following cases illustrate these points:

Case 1.—Hosp. No. 448034: C. C., male, age 30, had suffered ulcer symptoms for ten years He was operated upon for a perforation ten years ago. During the past six months he had vomited frequently and had lost weight. He had taken large doses of bicarbonate of soda the day of admission, October 31, 1930.

Physical examination disclosed generalized muscular rigidity, risus sardonicus, carpopedal spasm, and a positive Chvostek sign. The urine was alkaline, specific gravity

1.016, with a faint trace of albumin. Hemoglobin 63 per cent, carbon dioxide combining power 123.5 volumes per cent, chlorides, as sodium chloride, 340 mg. Blood urea 34 mg., calcium 8.5 mg., and phosphorus 5.8 mg. The gastric retention was 1,260 cc.

Course.—The patient was adequately treated with intravenous saline and glucose. He continued to vomit, and when treated with an indwelling tube, drained large amounts. Lavages did not materially decrease the gastric retention. The urinary specific gravity did not rise above 1.016. The blood carbon dioxide combining power and the chlorides approached normal limits but could not be kept there. It was felt that renal damage from a severe alkalosis, plus fluid and chloride loss from the stomach did not allow a more permanent correction of the alkalosis. It was, therefore, decided to perform a preliminary jejunostomy for alimentation. On November 10, 1939, exploration disclosed a huge, thin-walled stomach and a marked inflammatory reaction in the first portion of the duodenum. A typical Witzel jejunostomy was performed at a point 24 inches distal to the ligament of Treitz. Saline was given by drip into the

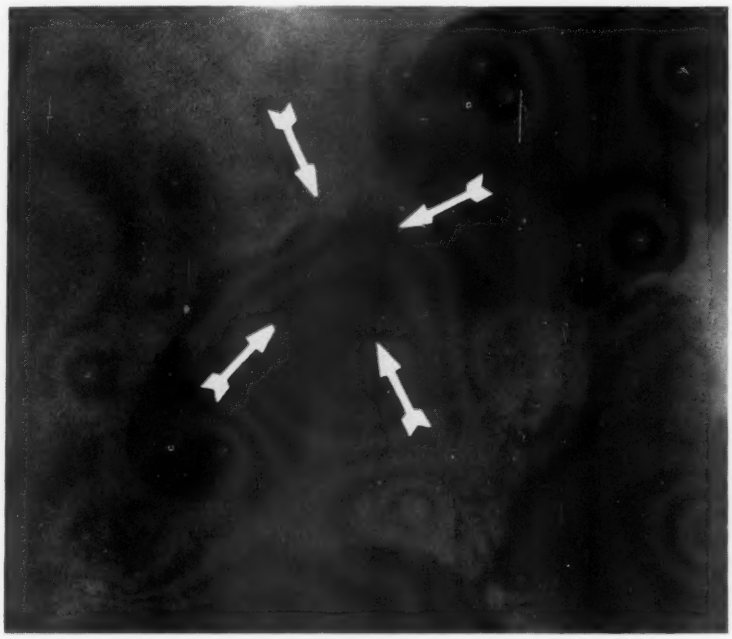


FIG. 1.—Case 2: Roentgenogram showing the large ulcer pocket.

jejunostomy tube and subsequently Scott-Ivy pabulum, vitamins, and Levin tube drainage in amounts up to 2,300 cc. daily. The patient had no cramps or diarrhea. He drained enormous amounts through the Levin tube at first. The intravenous drip which was being administered was stopped on the seventh postoperative day. The patient improved progressively. The blood chemistry figures returned to normal, and the urinary specific gravity rose to 1.020. He gained 13 pounds. By the eleventh postoperative day the gastric retention was only 30 cc. On the thirty-eighth postoperative day the patient was reoperated. A subtotal gastrectomy with an antecolic termino-lateral gastrojejunostomy of the Hofmeister type was performed. The jejunostomy was not disturbed. His postoperative course was remarkably uneventful. He was fed up to 1,800 cc. a day of Scott-Ivy pabulum. On the third and sixth day the patient had four diarrheal movements, which were controlled with small doses of deodorized tincture

of opium, administered through the jejunostomy. The tube was removed on the tenth day. The patient was discharged on the sixteenth postoperative day, very much improved.

He was seen in Follow-up Clinic, June 24, 1942, at wich time he had no gastric complaints. He had gained ten pounds since discharge.

This case illustrates the efficacy of a jejunostomy performed in a patient suffering from a complete pyloric stenosis with huge gastric retention, resulting in severe alkalosis and kidney damage.

Case 2.—Hosp. No. 485954: B. D., male, age 37, entered the hospital, February 20, 1942, complaining of recurrent epigastric pain, relieved by food, for the past seven years. He had lost 45 pounds during this time. He was rather emaciated. The roentgenograms disclosed a markedly deformed duodenal bulb with a definite ulcer pocket, and gastric retention. Hemoglobin 78 per cent, blood chlorides 515 mg. as sodium chloride, carbon dioxide combining power 91 volumes per cent. The gastric retention was 1,200 cc.

Course.—The patient was treated with intravenous glucose and saline. Lavages were given, and, in the interim, a Sippy diet and amphogel. The pain and alkalosis, nevertheless, continued. The patient was explored under spinal anesthesia February 26, 1942. A greatly dilated stomach was found. There was a large mass in the first portion of the duodenum, apparently a penetrating duodenal ulcer. A typical jejunostomy was performed. Postoperatively, the patient required parenteral fluids for three days only. Scott-Ivy pabulum and gastric drainage were given up to 2,000 cc. daily. Moderately severe cramps occurred on one occasion, but there was no diarrhea. The patient slowly improved, and as he retained a soft diet the jejunostomy feedings were diminished. The chemistry figures, however, never returned to normal. On the 74th postoperative day he was reoperated. The inflammation of the duodenum had decreased so that the swelling was one-third its original size (Figs. 1 and 2). A subtotal gas-

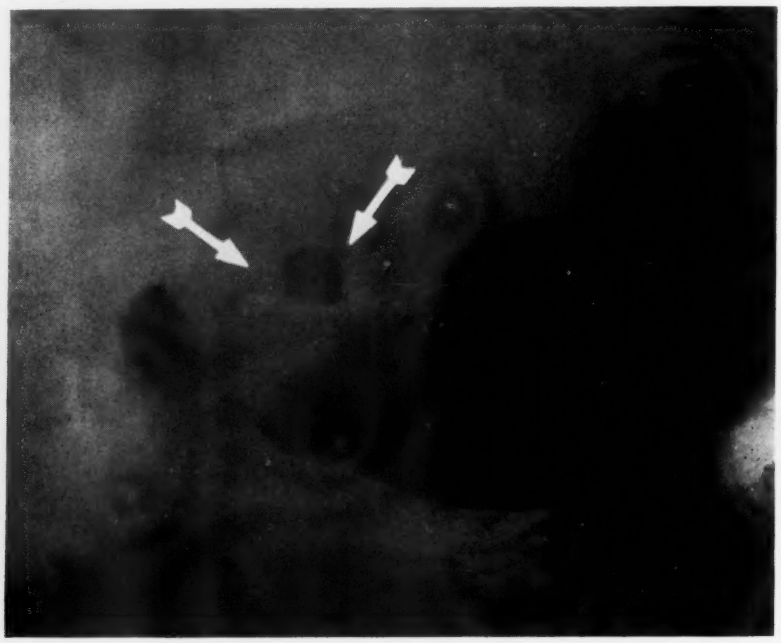


FIG. 2.—Case 2: Roentgenogram after ten weeks of jejunal alimentation. The ulcer pocket is one-third its former size.

trectomy with an antecolic anastomosis was performed. The postoperative course was uneventful. Intravenous therapy was discontinued after three days. Scott-Ivy pabulum and saline were administered in amounts up to 1,900 cc. daily. The tube was removed on the seventh day. He was discharged 19 days after his second operation.

He was seen in Follow-up Clinic July 22, 1942. The patient had no gastric complaints, and he had gained 20 pounds.

This case illustrates the advantage of jejunal alimentation in the control of intractable pain and alkalosis. It demonstrates quite graphically the partial subsidence of the local inflammatory reaction, and the diminution in the size of the stomach.

Case 3.—Hosp. No. 481601: F. G., male, age 44, had suffered pain, relieved by food, for five years. He had had a severe gastro-intestinal hemorrhage three years ago, and since then had milder attacks of bleeding at three- or four-month intervals. During the last five months the pain had become more severe, and was unrelieved by alkalis. In the past week the pain had increased, and he had noticed black stools. Vomiting was present for two days prior to admission November 10, 1941.

Physical examination disclosed a pale, emaciated male. There was moderate epigastric tenderness and spasm. The stool was tarry. Pulse 120; blood pressure 115/85. Hemoglobin 50 per cent. Blood urea 22 mg.; chlorides 515 mg.; carbon dioxide combining power 89 volumes per cent. He had a gastric retention of 1,900 cc.

Course.—Despite medical treatment and parenteral fluids, he continued to vomit and suffer pain. The bleeding stopped. The blood chemistries varied, and four weeks after admission, in spite of parenteral fluids, the blood carbon dioxide combining power was 105 volumes per cent, chlorides 455 mg., and urea 36 mg. Therefore, a typical preliminary jejunostomy was performed under spinal anesthesia December 9, 1942. Postoperatively, amounts of Scott-Ivy pabulum and gastric drainage were given up to 2,600 cc. daily. The blood chemistry, however, never reached normal. During a period of several weeks he was fed a Muelengracht diet and given atropine, but he continued to have occasional negative oral balances, and one episode of melena. With repeated transfusions the hemoglobin was brought up to 74 per cent. Exploration was performed under spinal anesthesia February 5, 1942. The stomach was twice normal size. Just beyond the pylorus was a marked constriction due to a posterior wall ulcer. A typical subtotal gastrectomy with an antecolic anastomosis was performed. The negative oral balances stopped after the fourth postoperative day. About 3,000 cc. of gastric aspiration and Scott-Ivy pabulum were given daily. Tube was removed on the fourteenth day. The patient was discharged on the sixteenth day.

He was seen in Follow-up Clinic four months later. He had no gastric complaints, and had gained 48 pounds.

This case illustrates the efficacy of enteral alimentation as opposed to parenteral hydration and alimentation in controlling a marked tendency toward alkalosis. It also discloses the fact that if a patient is properly prepared, the postoperative course for a long time after a bleeding episode will be uneventful.

Jejunostomy in these cases was always followed by a subtotal gastrectomy.

PALLIATIVE JEJUNOSTOMY

Jejunostomy as a definitive procedure in the surgery of gastroduodenal ulcer has outlived its usefulness. It has little to recommend it at present. It was only employed in those exceptional cases in which the general

physical condition of the patient and the characteristics of the local pathology permanently contraindicated any subsequent gastric surgery. The effects of palliative jejunostomy in these cases were similar to those obtained by the use of preliminary jejunostomy, but, inasmuch as further corrective surgery is prohibited, the clinical benefits will last only as long as the alimentation is continued. In one case (Hosp. No. 464135), jejunostomy was performed for an acute jejunal ulcer which developed after an extremely high subtotal gastrectomy and anterior vagotomy. Insufficient stomach remained for any further resection. The jejunal alimentation was continued for 96 days, and the patient improved. However, the ultimate prognosis is extremely doubtful. The second patient (Hosp. No. 482627) was a woman well advanced in years and afflicted with hypertension and cardiovascular disease. Following a gastro-enterostomy she developed a severe, bleeding, gastrojejunal ulcer which almost completely occluded the stoma and resulted in marked gastric retention with alkalosis. A palliative jejunostomy was performed 19 months ago, and the tube is still in place. Generally, she has improved. However, on occasions the ingestion of food causes episodes of gastric distress and bleeding. A return to the jejunal feedings relieves the symptoms. It is likely that if the tube is ever permanently removed there will be a return of the jejunal ulceration.

COMPLEMENTARY JEJUNOSTOMY

The largest number of jejunostomies for alimentation in this series has been the complementary type, those which were performed coincidentally with the gastric operation. This was done with the hope that it might neutralize many of the postoperative difficulties in gastric emptying. Some of the mechanisms involved in the pathologic physiology of atony of the stomach are not definitely known. Cases of acute gastric dilatation may be secondary to mechanical factors. Obstruction may be caused by an occluding edema of the gastro-enteric stoma which is often inflammatory at first, and subsequently is abetted by the biochemical disturbances incident to hypoproteinemia.[8, 11] Occasionally an acute obstruction may develop in a retrocolic gastro-enterostomy because of a prolapse of the stoma or the efferent loop into the lesser omental sac. In a few cases, the postoperative development of a plastic exudate and adhesions may be sufficient to occlude either the afferent or efferent jejunum. However, there are cases of acute gastric dilatation or atony in which neither operative exploration nor postmortem examination reveals any anatomic cause for the clinical symptomatology of obstruction. This type of gastric ileus has been attributed to many factors. Any operative procedure upon the stomach, regardless of the technical skill of the surgeon, is accompanied by a certain amount of unavoidable trauma often causing muscular paresis and atony. In addition, a subtotal gastrectomy which entails the excision of a greater part of the lesser curvature and partial destruction of the gastrohepatic omentum causes considerable

damage to the vagus nerve. This adversely affects the motor activity of the stomach. Moreover, in those cases in which the stomach has been previously dilated, there is no reason to assume that any anastomotic procedure or radical gastric resection will immediately remedy the atony. It is a common experience following a gastro-enterostomy or a gastric resection that 24 to 48 hours may elapse before the stomach partially regains its peristaltic activity. The inevitable sequela is the stagnation of gastric contents causing repeated vomiting, or persistent gastric drainage through an indwelling Levin tube.

Fortunately, however, in the majority of patients these difficulties in gastric emptying are transient and inconsequential. If they persist, they portend serious consequences. The daily loss by vomiting or gastric drainage of several thousand cubic centimeters of ingested fluids, gastric and duodenal secretions, rapidly causes dehydration, inanition, avitaminosis, hypoproteinemia and alkalosis. These chronically ill individuals need adequate and proper nourishment from the very beginning of their postoperative period. This is not completely supplied in some patients by the parenteral administration of blood, plasma, vitamins, amino-acids and electrolytes. It is the systemic effects produced by gastric atony in its varying severity and duration which adds to the morbidity and mortality of gastric surgery.

Inasmuch, as gastric atony is always a potential postoperative complication, prophylactic operations have been devised to eliminate its consequences. These procedures have been reviewed in detail by Perman.[10] Any method to be effective in the treatment of gastric atony must fulfill certain requirements. It must keep the dilated stomach decompressed, it must provide adequate fluid and nourishment during this period of gastric inactivity, and it must prevent alkalosis. It would seem that the two-way gastro-enterostomy tube developed by Abbott and Rawson[1] would satisfy these desiderata. However, the use of this tube in our experience has not been satisfactory. Its introduction from the stomach into the efferent jejunal loop during a subtotal gastrectomy is not an easy procedure, especially if asepsis is to be maintained. Moreover, during the postoperative period it may be inadvertently dislodged either by an uncooperative patient or through carelessness. Furthermore, these tubes cannot be kept in place indefinitely because of the danger of damage to the arytenoid cartilages. We have seen two cases in which an edema of the larynx developed from an arytenoiditis. If the two-way tube were entirely satisfactory in the decompression of the stomach and in the simultaneous maintenance of nourishment, jejunostomy for alimentation would not have been recently revived in gastric surgery. This is evidenced by the papers of Wesson,[14] Allen and Welch,[2] and Clute and Bell.[3]

In 1929, Kirschner[9] advocated the complementary use of jejunostomy in gastroduodenal surgery, and he utilized this procedure in 60 cases of subtotal gastrectomy. He reported that it was extremely useful. It supplied immediate nourishment to those who had suffered so long from an ulcer that

their capital resources had already been drawn upon for their sustenance. Moreover, the deleterious and dire consequences of a gastric ileus was often neutralized by the complementary jejunostomy.

The importance and the practical applications of Kirschner's contribution evidently have not been fully appreciated. Since 1938, we have used complementary jejunostomy in an increasing number of patients with gratifying results. Indications have been gradually established so that it is now employed routinely in certain types of cases. It was used in those gastric and duodenal ulcers which were complicated by stenosis and marked gastric dilatation, for, as has been stated previously, a stomach which has been atonic prior to operation is likely to be atonic following operation. Complementary jejunostomy was especially helpful in those instances in which the technical procedures were unusually traumatic or complicated, as in extensive resections for gastrojejunal ulcers or those at the cardia. These patients were found to be especially prone to gastric ileus. Another indication for the use of concomitant jejunostomy was found in those cases in which a dehiscence of a friable duodenal stump was a possibility. These were drained routinely, and if a fistula developed the duodenal contents were aspirated by suction and fed together with the pabulum *via* the jejunostomy. In this way the nutrition and normal chemical balances were usually maintained. Complementary jejunostomy has been performed in 38 cases. It was used in 11 cases of gastro-enterostomy and in 21 of subtotal gastrectomy. In four instances it was established simultaneously with a resection of the stomach following either a previous gastrectomy or a gastroenterostomy. In one case it was performed as a coincident procedure to the restoration in continuity for a gastrojejunocolic fistula, and in another patient it was used as an aid to an entero-anastomosis for a stenosing ulcer of the afferent jejunum.[5] It is undoubtedly true that complementary jejunostomy was unnecessary in some cases, as judged by the subsequent postoperative course. However, even in these patients it did no harm and certainly simplified the postoperative alimentation and diminished considerably the period of time in which intravenous therapy was required.

The following cases, which are abstracted, demonstrate the efficacy of complementary jejunostomy:

Case 4.—Hosp. No. 457630: I. S., male, age 58, was admitted, June 1, 1940, because of epigastric pain, eructations and vomiting of 25 years duration. Examination disclosed an edentulous male. There was tenderness in the upper abdomen, and a definite succussion splash could be elicited. The hemoglobin was 84 per cent, blood chlorides 410 mg., and carbon dioxide combining power 74 volumes per cent. There was 2,200 cc. of retained food and fluid in the stomach. Roentgenologic examination showed a marked deformity of the duodenal bulb and an ulcer pocket on the lesser curvature side. There was a delay in gastric emptying. Thirty-five per cent of the barium was retained after six hours.

The patient was prepared for operation with intravenos saline and glucose, lavages, and vitamins, for 16 days. On June 17, 1940, exploration, under cyclopropane anesthesia, disclosed a large duodenal ulcer perforating into the head of the pancreas. A typical subtotal resection and jejunostomy for alimentation was performed. Post-

operatively, the patient developed a typical gastric atony, which persisted for 16 days. During this time alimentation was maintained by feeding Scott-Ivy pabulum and Levin tube drainage through the jejunostomy in amounts up to 2,500 cc. daily, augmented by intravenous saline and glucose and transfusions. On the eleventh postoperative day roentgenograms disclosed complete retention of barium in the stomach after 25 hours (Fig. 3). The atony subsided after the sixteenth day and the patient improved

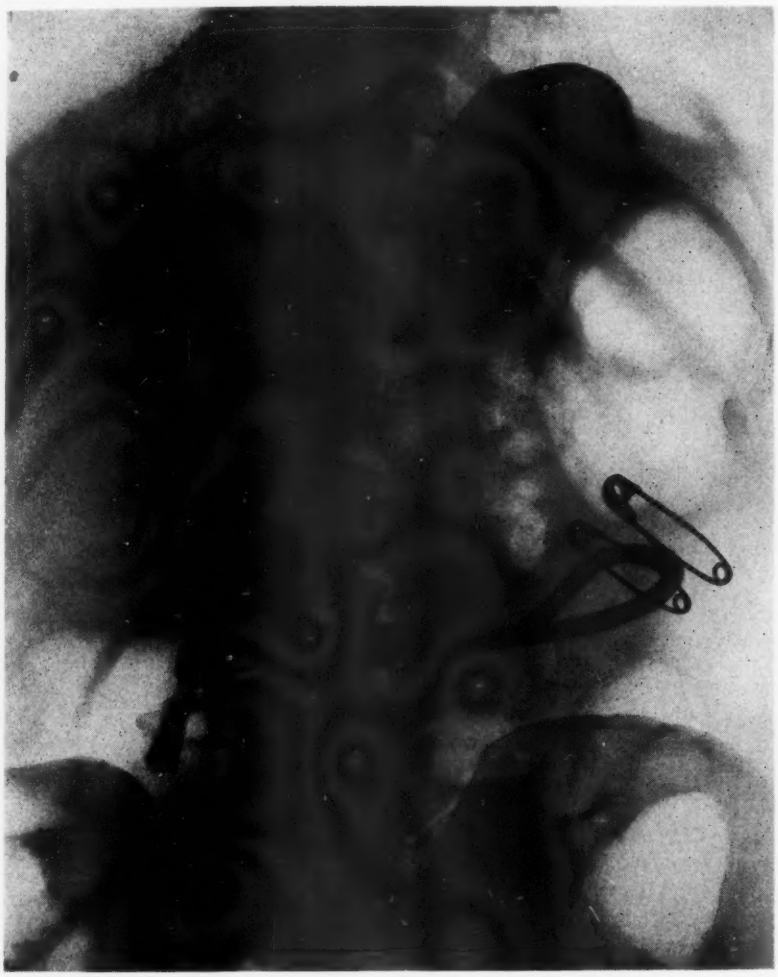


FIG. 3.—Case 4: Roentgenogram taken on eleventh postoperative day, 25 hours after the administration of barium. Note that none has left the stomach. The jejunostomy tube is seen *in situ*.

rapidly. On the twenty-fifth day roentgenograms disclosed a normally emptying stomach (Fig. 4). The tube was removed on the twenty-second day and he was discharged on the twenty-sixth postoperative day.

This case illustrated the fact that preoperative gastric atony is not relieved by resection, but rather that the ileus continues. The importance and convenience of a jejunostomy for alimentation is readily appreciable.

Case 5.—Hosp. No. 453628: G. C., male, age 72, had originally entered the hospital because of recurrent indigestion, epigastric pain, vomiting and bleeding, in August,

1939. Roentgenograms disclosed a deformed bulb with marked delay in emptying time. He was transfused several times. His blood urea rose to 118 mg., and the specific gravity of the urine remained low, about 1.006. It was felt that these changes represented kidney damage due to alkalosis incident to the vomiting and prolonged ingestion of bicarbonate of soda. He was discharged much improved, only to be readmitted several months later with an acute perforation of his ulcer. This was sutured, and the patient recovered. On May 12, 1940, he was readmitted because of weakness, anorexia, and burning pain in the epigastrium. Examination disclosed an eldery, emaciated male

FIG. 4.—Case 4: Roentgenogram taken on twenty-fifth postoperative day disclosing a normally emptying stomach.

deaf-mute. There was general epigastric tenderness and evidences of marked arteriosclerosis. Hemoglobin was 49 per cent. Blood carbon dioxide combining power 63.1 volumes per cent, and chlorides 595 mg. There were 2,000 cc. of gastric retention. The patient was transfused several times, and was lavaged frequently. He continued to have many attacks of epigastric pain, some of which were relieved by removing large quantities of gastric retention.

Course.—On April 15, 1940, under local anesthesia, the patient was explored. The stomach was found to be twice normal size, and there were multiple adhesions in the region of the duodenum. In the first portion of the duodenum there was a scar

and contraction due to the duodenal ulceration. A typical posterior gastro-enterostomy was performed, and a tube-jejunostomy established 12 inches below the anastomosis. Postoperatively, the patient had large negative oral balances for 12 days. He was fed Scott-Ivy pabulum in increasing doses up to 2,880 cc. daily, without diarrhea or cramps. An intravenous was necessary for four days only. His blood chemistry values remained normal. The urinary output was good. The tube was removed on the nineteenth postoperative day, and the wound healed promptly.

He was seen in the Follow-up Clinic, July 22, 1942, at which time he had gained 25 pounds, and had suffered no gastric complaints.

This case is typical of the excellent results obtained from the establishment of a jejunostomy for alimentation in a seriously ill, elderly male, who had suffered long-continued gastric dilatation due to obstruction, and in whom it was to be expected that a postoperative gastric ileus would develop. It is typical of the advantages of the complementary jejunostomy.

SHORT ABSTRACTS

Case 6.—Hosp. No. 454659: B. L., male, age 40, had suffered peptic ulcer disease for six years, and had been repeatedly admitted to the hospital for pain, vomiting, perforation and alkalosis. At operation, his stomach was found three times normal size. A subtotal resection and jejunostomy for alimentation were performed. Postoperatively, his course was uneventful except that he had persistent negative oral balances, as high as 2,150 cc. daily. He was fed large amounts, up to 3,450 cc., of Scott-Ivy pabulum daily. The negative oral balance continued for nine days. The tube was removed on the thirteenth day, and the patient discharged on the fourteenth day.

Case 7.—Hosp. No. 468543: H. H., male, age 41, had suffered ulcer symptoms, including one episode of perforation, for 13 years. He entered the hospital after several months of vomiting. Gastric retention was 2,100 cc. At operation, an enormous dilatation and hypertrophy of the stomach, and a large inflamed duodenal ulcer were seen. A typical subtotal resection and jejunostomy were performed. The patient continued, postoperatively, to have negative oral balances for 14 days. He was fed up to 1,700 cc. of pabulum daily, and this, as well as intravenous saline, glucose, and amino-acids, did much to help him through the prolonged period of gastric atony.

Case 8.—Hosp. No. 480134: S. B., male, age 41, entered the hospital for surgical treatment of a penetrating jejunal ulcer. A secondary subtotal resection and a jejunostomy were performed. The patient had a negative oral balance for nine days and was fed pabulum, Levin tube drainage, vitamins and sulpha drugs through the jejunostomy tube. Despite a rather severe postoperative pneumonia, he recovered.

Case 9.—Hosp. No. 465396: A K., male, age 40, upon whom a high subtotal resection, with a Murphy button anastomosis, was performed because of a highly situated lesser curvature ulcer. Postoperatively he had a negative oral balance for 11 days, and was tided through this difficult period by large amounts of jejunostomy feedings and replacement of Levin tube drainage.

Even in this small series, it becomes quite apparent that if a concomitant jejunostomy had not been performed, a supplementary jejunostomy would probably have been necessary in some cases because of a persistent gastric ileus. These cases, from reports in the literature, are always attended by an appreciable mortality.

SUPPLEMENTARY JEJUNOSTOMY

Naturally, there are cases in which the possible occurrence of a gastric atony following operative procedures upon the stomach cannot be predicted.

Severe gastric atony in some patients may occur from the very beginning; in others it may not become evident until a week or ten days after operation. The clinical picture, however, is similar in both groups of cases. It is recognized by the symptoms and the physical findings of a high intestinal obstruction, in which progressive deterioration occurs in spite of adequate parenteral treatment. There is always a tendency to procrastinate too long with conservative therapy, reserving operative intervention until it is too late. This is undoubtedly the reason that the results of secondary procedures in these cases are attended by such a prohibitive mortality. When gastric ileus was suspected because alkalosis and increasing or stationary large negative oral balances were present, roentgenologic examination of the stomach with barium proved to be invaluable and indispensable. It revealed a dilated stomach in which the barium was completely retained, or it disclosed other reasons for the obstruction, as, for example, an occlusion of the efferent jejunum. We agree with the conclusions of Allen and Welch,[2] that in the older-age group of patients exploration should be undertaken after seven days if the amounts obtained by gastric aspiration show a stationary or progressive increase and the chemical evidences of alkalosis are present. In younger patients conservative measures may be tried for a longer period of time.

Many operative measures have been advocated to correct or relieve the effects of gastric atony. Surgeons have advised another gastro-enterostomy when a malfunction of the previous one was suspected. This usually is futile. Entero-enterostomy has been suggested if there is a marked dilatation of the afferent loop in contrast to a contraction of the efferent jejunum. This rarely relieves gastric atony and, moreover, it subsequently deprives the stomach of the full benefits of duodenal alkalinization. Hoag and Saunders[7] reported several cases in which a gastric ileus was relieved by jejunoplasty performed in the region of a malfunctioning gastro-enteric stoma. Many Continental surgeons have been enthusiastic in their praise of a tube-gastrostomy. However, similar benefits are obtained by the routine postoperative use of continuous suction drainage of the stomach.

Supplementary jejunostomy for alimentation in cases of gastric atony was suggested at a very early period in the development of gastric surgery.[13] In our experience, it has proven to be most satisfactory. It is a simple technical procedure which can be performed through the original operative incision expeditiously and without shock. To repeat, it is a method which, when combined with the concomitant use of gastric drainage, decompresses the stomach, maintains nutrition, and corrects alkalosis.

The following cases are of interest:

Case 10.—Hosp. No. 467016: J. M., male, age 25, was admitted, December 27, 1941, complaining of epigastric pain, relieved by food or soda, of five years duration. Two years ago he had an episode of bleeding, and one year ago a perforation of a duodenal ulcer was sutured. Three days prior to admission he noticed a tarry stool, and subsequently fainted. He had had poliomyelitis in childhood, and had been left with a

partial paralysis of the right upper and both lower extremities, despite numerous corrective operations.

Physical examination disclosed a pale young male. There was moderate epigastric tenderness. The hemoglobin was 52 per cent, and the blood chemistries were normal.

Course.—The patient was transfused repeatedly. The stools became guaiac-negative, and the hemoglobin rose to 75 per cent. On January 15, 1942, under spinal anesthesia, a subtotal gastrectomy with antecolic anastomosis was performed for an active duodenal ulcer with stenosis. Postoperatively, from the beginning the Levin tube drained large amounts. Despite adequate parenteral therapy, alkalosis developed. The blood carbon dioxide volume power rose to 83.2 volumes per cent, and the blood chlorides to 540 mg. prior to operation. On February 2, 1942, under spinal anesthesia, the median incision was reopened and the operative site examined. No organic cause for obstruction was found. A purse-string jejunostomy was performed. Postoperatively, the patient developed an active phlebitis of the dorsum of the right hand, and suffered chills and high fever. Blood culture was positive for a gram-negative bacillus. The sepsis subsided with chemotherapy. He continned to vomit. On the seventh postoperative day blood chemistries showed carbon dioxide combining power 58.4 volumes per cent, chlorides 620 mg., urea 9 mg. Through the jejunostomy he was given up to 3,200 cc. of Scott-Ivy pabulum, gastric drainage, saline and sulfathiazole for 18 days. One mild episode of diarrhea was controlled by the administration of tincture of opium. The patient had a negative oral balance for 13 days after jejunostomy (a total of 31 days after the original gastrectomy). After the eighteenth day, alimentation was entirely oral. The jejunostomy tube was removed on the twenty-fourth day, and he was discharged.

He was seen in the Follow-up Clinic, October 28, 1942, at which time he was in excellent health.

This patient also illustrates the value of the supplementary jejunostomy for a gastric ileus, which persisted 31 days after gastrectomy. Because of a marked phlebitis and sepsis, parenteral therapy was impossible, and jejunostomy alone controlled the alkalosis and maintained nutrition.

Case 11.—Hosp. No. 484235: D. W., male, age 50. Thirteen years ago, following an episode of malena, a perforation of a duodenal ulcer occurred. This was sutured, and since then he has suffered epigastric pain relieved by food and soda. Roentgenologic examination on admission disclosed a large penetrating duodenal ulcer. The blood chemistries were normal.

Course.—Exploration, under spinal anesthesia, January 19, 1942, disclosed a dilated stomach, with a marked deformity of the duodenum and a small diverticulum. A typical subtotal gastrectomy with antecolic anastomosis was performed. Postoperatively, from the beginning, the patient drained large amounts through the Levin tube, and vomited when it was removed. Hydration was maintained by the intravenous route. The carbon dioxide combining power rose to 84 volumes per cent by the fifth postoperative day. Inasmuch, as the negative oral balance varied between 2,500 and 3,000 cc. per day, exploration was deferred until the seventh postoperative day.

On January 26, 1942, under spinal anesthesia, and through a left rectus incision, the operative site was examined and found to be normal. Eighteen inches distal from the anastomosis a typical Witzel jejunostomy was performed. The patient was subsequently fed up to 88 ounces daily of Scott-Ivy pabulum, vomitus, Levin tube drainage and vitamins. The carbon dioxide combining power dropped to 59.2 volumes per cent on the fourth postoperative day. He developed a right upper lobe pneumonia, which responded to chemotherapy. A dehiscence of the left rectus incision was treated by packing.

This patient continued to have a negative oral balance for 13 days following the supplementary procedure. During this period alimentation was maintained by jeju-

nostomy which, together with intravenous amino-acid, kept the blood proteins within normal limits. Twenty-five days after the second operation the jejunostomy tube was removed. The wound healed rapidly, and he was discharged February 22, 1942.

He was seen in Follow-up Clinic, April 22, 1942, at which time he had no gastric complaints, and felt quite well.

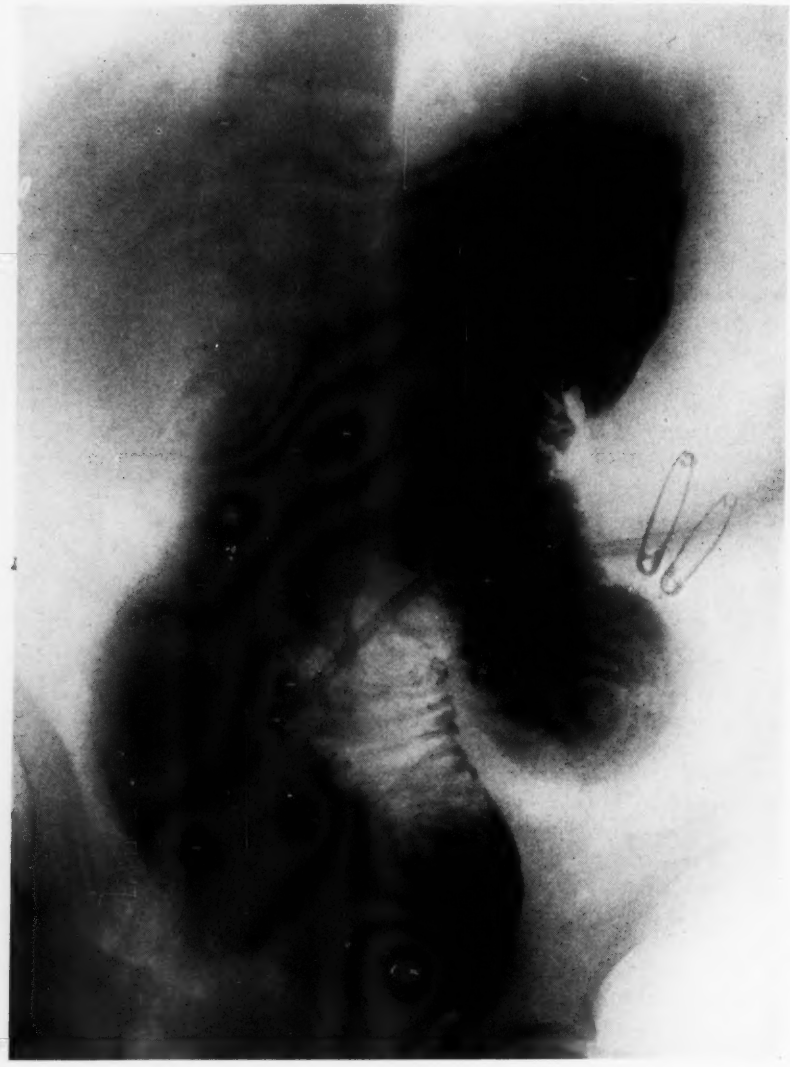


FIG. 5.—Roentgenogram disclosing complete obstruction at site of a Witzel jejunostomy.

This case illustrates the proper employment of the supplementary jejunostomy. When the gastric ileus showed no signs of abating, and alkalosis progressed despite intravenous therapy, the patient, who was age 50, was promptly explored. No organic cause for the obstruction was found. A jejunostomy was performed with excellent results.

THE TECHNIC OF JEJUNOSTOMY AND JEJUNAL ALIMENTATION

The history and clinical application of jejunostomy for alimentation has been thoroughly and adequately reviewed by Wolfer.[15] The technic employed for jejunostomy in this clinic was quite simple, and added but a few moments to the operative time, when performed coincidentally with a gastric procedure. If the jejunostomy was preliminary, a median epigastric incision was made from the ensiform to the umbilicus, under spinal or local anesthesia. This exposure afforded ample opportunity to explore the upper abdomen and gave easy access to the identification of the duodenojejunal angle. The intestine was traced distally for about 18 to 24 inches from the ligament of Treitz. If a loop of bowel is picked at random in the left upper quadrant on the assumption that it is high jejunum, the operator may be dismayed when he subsequently finds, at postmortem examination, that he has performed a low ileostomy.

The Witzel type of jejunostomy was performed in the first 39 cases. There were two complications, one, an instance of acute intestinal obstruction which developed at the site of the jejunostomy and necessitated operative intervention (Fig. 5.) (Hosp. No. 480550), and in the other patient the signs of local peritonitis developed in the region of the enterostomy. This subsided with the removal of the tube.

Recently, impressed by the experiences of others,[2, 3] we have performed the operation in 12 cases according to the Stamm principle. Through a small opening in the jejunum, a No. 12 to 14 F. whistle-tip catheter was fed aborally for about six inches. The tube was then fixed to the bowel with catgut and was buried by means of two purse-string linen sutures. The patency of the jejunostomy was tested immediately by the introduction of saline. The catheter was simply brought through a stab wound in the left subcostal region and attached to the skin with silk.

Four hours after operation 60 cc. of warm saline was introduced, and this was repeated every two hours. After 12 hours 60 cc. of the Scott-Ivy[12] pabulum, alternating with saline, were given regularly. The amount was gradually increased to 120 cc., or more, hourly. The Scott-Ivy pabulum, which is usually well tolerated by the jejunum, is a nonirritating mixture of high caloric content and necessary food elements. The feedings, which are warmed, are given slowly, preferably by gravity drip, or by syringe. If given too rapidly or in too large quantities, jejunal regurgitation may occur. This is easily recognized because, if suspected, insoluble carmine may be mixed with the pabulum and recovered in the gastric drainage. In about ten per cent of the cases, these feedings caused diarrhea and cramps, which were usually controlled by appropriate doses of tincture of opium and bismuth subcarbonate. Amino-acids and medications have also been given by jejunostomy. The tube occasionally became plugged, but the obstruction was relieved by irrigation with a ureteral catheter. When the loss of gastric contents through the indwelling tube was appreciable,

the drainage was collected and part of it was introduced through the jejunostomy, alternating with the pabulum feedings. Jejunal alimentation was continued until the stomach emptied normally. The tube was removed, as a rule, 48 hours after the jejunal feedings had been discontinued. The earliest removal in the complementary cases was nine days, the latest 35 days, and the average 15 days. Although there was some drainage of bile for 24 hours, or more, after its removal, the discharge soon ceased spontaneously in both the Witzel and Stamm types of jejunostomy.

Jejunostomy *per se* did not account for a single mortality in this series.

SUMMARY

1. Jejunostomy for alimentation was performed in a series of 51 patients suffering from recurrent chronic peptic ulcer.

2. It was used as a preliminary, palliative, complementary, and supplementary measure in the surgery of gastroduodenal ulceration. The indications and clinical applications of these are fully described.

3. The technic of jejunostomy and jejunal alimentation is discussed.

4. Evaluation of results obtained.

REFERENCES

1 Abbott, W. O., and Rawson, A. J.: A Tube for Use in Postoperative Care of Gastroenterostomy Patients. J. A. M. A., **112**, 2414, 1939.

2 Allen, A. W., and Welch, C. E.: Jejunostomy for the Relief of Malfunctioning Gastro-enterostomy Stoma. Surgery, **9**, 163, 1941.

3 Clute, H. M., and Bell., L. M.: Jejunostomy for Postoperative Feeding. ANNALS OF SURGERY, **114**, No. 3, 402, 1941.

4 Coller, F. A., Bartlett, R. M., Bingham, L. C., Maddock, W. G., and Pedersen, S.: The Replacement of Sodium Chloride in Surgical Patients. ANNALS OF SURGERY, **108**, 769, 1938.

5 Colp, R.: Surgical Problems in the Treatment of Gastrojejunal Ulceration. ANNALS OF SURGERY, **114**, 543, October, 1941.

6 Eusterman, G. B., and Balfour, D. C.: The Stomach and Duodenum. W. B. Saunders, Philadelphia, 1936.

7 Hoag, C. L., and Saunders, J. B.: Jejunoplasty for Obstruction Following Gastroenterostomy or Subtotal Gastric Resection. Surg. Gynec. and Obst., **68**, 703, 1939.

8 Jones, C. M., and Eaton, F. B.: Postoperative Nutritional Edema. Arch. Surg., **27**, 159, 1933.

9 Kirschner, M.: Die Prophylactische Jejunostomie bei Magenoperationen. Arch. f. klin. Chir., **157**, 561, 1929.

10 Perman, E.: Surgical Treatment of Gastric and Duodenal Ulcer. Acta. Chir. Scand., **77**, suppl. 38.

11 Ravdin, I. S.: Factors Involved in the Retardation of Gastric Emptying after Gastric Operations. Penn. M. J., **41**, 695, 1938.

12 Scott, H. G., and Ivy, A. C.: Jejunal Alimentation. ANNALS OF SURGERY, **93**, 1197, 1931.

13 Stieda, A.: Über die Vorbereitung und Nachbehandlung bei Magenoperationen. Arch. f. klin. Chir., **63**, 715, 1901.

14 Wesson, H. R.: Postoperative Gastric Retention Treated by Jejunostomy for Feeding Proc. Mayo Clin., **12**, 727, 1937.

15 Wolfer, J. A.: Jejunostomy with Jejunal Alimentation. ANNALS OF SURGERY, **101**, 708, 1935.

DIVERTICULA AND VARIATIONS OF THE DUODENUM

WOLFGANG ACKERMANN, M. D.

NEW YORK, N. Y.

FROM THE DEPARTMENT OF SURGERY, COLLEGE OF PHYSICIANS AND SURGEONS, COLUMBIA UNIVERSITY, NEW YORK, N. Y.

IN THE REGIONAL ANATOMY COURSE given to third year medical students at the College of Physicians and Surgeons no difficulty was encountered in demonstrating anomalies of the abdominal viscera except as to diverticula of the duodenum. It seemed reasonable to assume that such abnormalities might elude observation, especially when they are small or embedded in the pancreatic tissues. To overcome this obstacle the idea was advanced that making casts of the duodenum would be a reliable method of demonstrating them.

The ideal material for this purpose appeared to be "Korogel," a molding jelly. However, because of certain technical difficulties connected with the handling of it, plaster of paris was used instead, and it proved entirely satisfactory.

A few experiments were first made on cadavers in the Departments of Anatomy and Pathology. Then, with the beginning of the school year 1938–1939 and through 1940–1941, every cadaver that came to the laboratory for use by Dr. Allen O. Whipple in teaching surgical anatomy, was examined for the presence of duodenal diverticula. During that period 50 bodies were studied. They were not selected, but taken at random.

PROCEDURE: When the abdomen is opened and the study of the viscera is about to begin, the omentum with the transverse colon and stomach are lifted up and turned back. The pylorus is mobilized, a piece of cord is passed underneath and tied snugly around it. The duodenum is left *in situ*. The duodenojejunal junction is located and a piece of cord is passed beneath through the mesentery. The jejunum is incised transversely on its antimesenteric border, about one inch below its junction, just enough to permit the insertion of an adapter to an all-metal 250 cc. syringe. The jejunum is now tied once over the adapter with the cord placed beneath it. The syringe is filled with plaster, which is injected slowly, exerting only sufficient pressure to ensure its entering any recess or diverticulum that may be present.

When the duodenum is filled to its capacity, the plaster commences to flow back. Then, as the syringe with its adapter is being quickly removed, an assistant ties a second knot to prevent the reflux.

After the plaster hardens sufficiently, the common duct is probed for its patency and then injected with about two to three centimeters of methylene blue, to establish the relation of the ampulla of Vater to the duodenum.

The following day the duodenal wall is incised along its entire convex

border and the model is removed with great care, while both the interior of the duodenum and the cast are being studied.

It is not within the scope of this paper to discuss, in detail, classification, etiology, age and sex incidence, size, location, distribution, *etc.*, of duodenal diverticula. With regard to such particulars the reader is referred to the literature. However, it must be pointed out that all diverticula herein described belong to the *primary* group, meaning those which have no obvious cause for their appearance, in contrast to those which are produced for instance by ulcers or adhesions and are called *secondary, true,* or *acquired.*

The *primary* are also called *false* or *congenital.* Although not proven, they are congenital in their nature, as they represent abortive attempts at the formation of a supernumerary pancreas. The pancreatic anlages cause local defects in the musculature and the possibility of pouching results from age, intestinal atony or an increase of intraduodenal pressure. This view is the opinion of Letulle, Tandler, Falconer, Lewis and Thyng.

Illustrations of 11 specimens (Figs. 1–11) among a total of 50 show single or multiple diverticula. This is *22* per cent, the highest figure so far recorded.

Although Grant is of the opinion that percentages convey little if any information, unless the statement bears with it reference to age, this is believed to be true only from the theoretic point of view. Clinically speaking, one must consider duodenal diverticula in differential diagnosis regardless of the age, since they occur at all ages.

The total number of the diverticula found is 14, if one bilocular (*a* and *b* of Fig. 2), meaning a combination of two diverticula with a common orifice, *c*, is accepted as a single diverticulum.

Of the 11 specimens with diverticula, eight (Figs. 1, 3, 5, 6, 7, 8, 9 and 11) had one, and three (Figs. 2, 4 and 10) had two diverticula.

The distribution is as follows: Five diverticula in the second part of the duodenum (*a–c*, of Fig. 2, Fig 3, *a* and *b* of Fig. 4, and Fig. 5); one between the second and third parts (Fig. 6); five in the third part (*d* of Fig. 2, Figs. 7, 8, 9 and *a* of Fig. 10); and three in the fourth (Fig. 1, *b* of Fig. 10, and Fig. 11). Thus, the opinion of practically all writers that the greatest number of diverticula occur in the second part cannot be corroborated by the findings recorded in this paper.

No diverticula were found in the first part; and all sprang from the concave, pancreatic border, which is morphologically the mesenteric border of the duodenum. All but four (*a–c* of Fig. 2, *d* of Fig. 2, Fig. 3 and *a* of Fig. 4) were buried in the substance of the pancreas, and had they not been filled with plaster a number of them would certainly have escaped detection.

As to their shape, it is obvious that the greater number are globular; some are funnel-shaped and some cylindrical.

The dimensions of the diverticula given in the Figures are probably

FIG. 1.

FIG. 2.

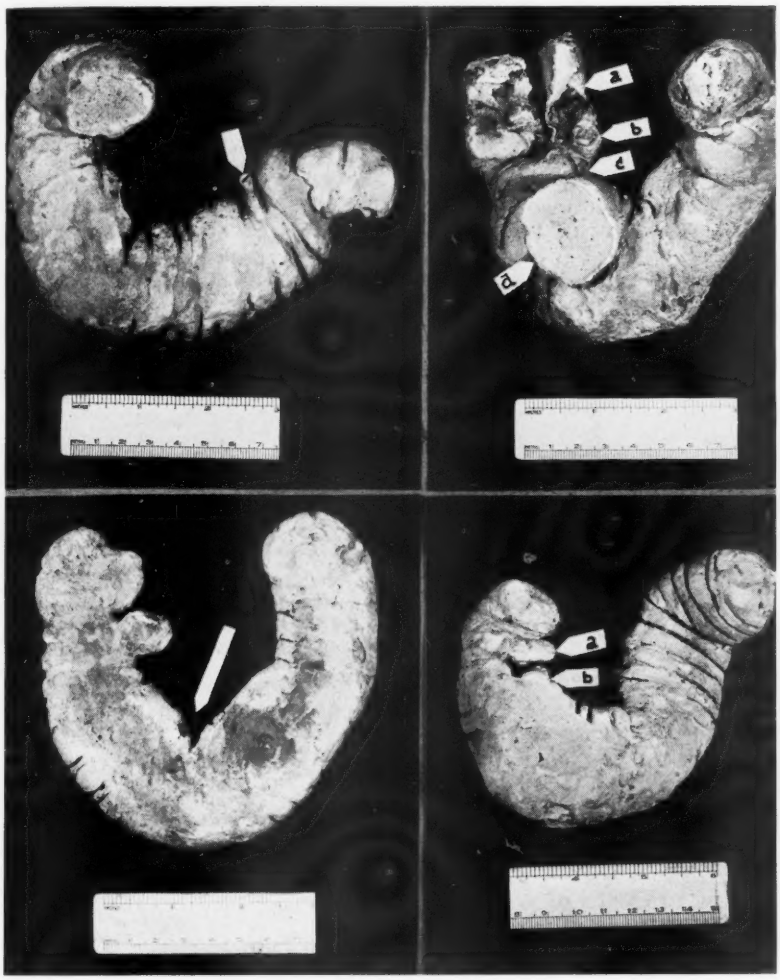


FIG. 3.

FIG. 4.

FIG. 1.—Arrow points to a small diverticulum in the fourth part of the duodenum, the smallest in the series.

FIG. 2.—The specimen shows a bilocular diverticulum *a* and *b*, arising from a common orifice *c* in the second part. Diverticulum *b* projects anteriorly and is, therefore, not demonstrated distinctly in the photograph. A second diverticulum is seen at *d*, in the third part of the duodenum. It is large, globular, and like the bilocular it arises on the pancreatic border; anteriorly, it appears to have a flat, smooth surface, a condition caused by the presence of air at the time of casting it.

FIG. 3.—A single diverticulum of the second part has a fairly large orifice. The arrow points to the location of the papilla of Vater.

FIG. 4.—Two diverticula are seen, both in the second part; diverticulum *a* extends medially and anteriorly, and diverticulum *b* posteriorly.

FIG. 5. FIG. 6.

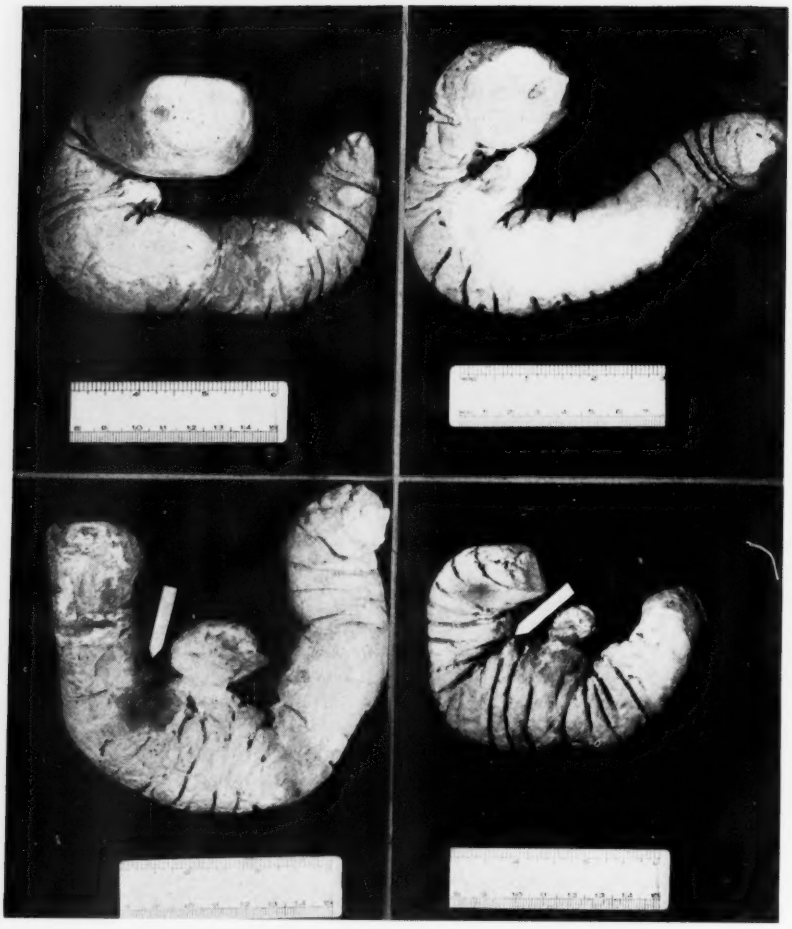


FIG. 7. FIG. 8.

FIG. 5.—The first part of the duodenum is large, the second and fourth parts are short. A single diverticulum may be seen at the lower end of the second part.

FIG. 6.—A fairly large diverticulum with a broad opening appears at the junction of the second and third parts of the duodenum.

FIG. 7.—The diverticulum in the third part of the duodenum is large and has a narrow orifice. The arrow points to the location of the papilla of Vater.

FIG. 8.—This is the smallest duodenal cast in the series. The diverticulum in the third part has a narrow orifice. The arrow indicates the location of the papilla of Vater.

smaller than those during life because of some shrinkage due to the embalming process; nevertheless, it is believed that the difference is of no practical importance.

Before roentgenologic examination became available, and made early recognition of this interesting lesion possible, during a period of 212 years (1710–1912), less than 100 cases were reported, and they were of only academic interest. They were found at necropsies exclusively.

How little attention they received in those days is evident from Cruveilhier's report in 1849. When writing on diverticula of the gastro-intestinal tract, he described pouches in the esophagus and colon, and stated that diverticula did not occur between these points.

From the moment when Case first diagnosed a duodenal diverticulum roentgenographically, in 1913, the number of reported cases commenced to increase rapidly, and, in 1932, Lockwood was able to cite a grand total of 357 cases diagnosed roentgenologically.

This total compared with the one recorded for the preroentgenographic period is high, and especially so when taking into consideration the great difference in the length of the two respective periods. Relatively speaking, however, no actual increase of duodenal diverticula was observed.

That this is true can be seen when roentgenologic figures are compared with the figures obtained at necropsies and both are expressed in percentages, as demonstrated in Table I. Indeed, a much higher frequency is observed in autopsy reports.

TABLE I

PERCENTAGE-FREQUENCY OF DUODENAL DIVERTICULA FOUND AT AUTOPSIES AND AT ROENTGENOLOGIC EXAMINATIONS

Author	Year	Number of Autopsies	Number of x-ray Examinations	Duodena with Diverticula	Percentage
Schuppel	1880	45		7	15.5
Rosenthal	1908	100		3	3
Baldwin	1911	105		14	13.3
Linsmayer	1914	1367		45	3.3
Horton & Mueller	1933	216		11	5.09
Grant	1935	133		15	11.3
Case	1920		6847	85	1.2
Andrews	1921		2200	26	1.18
Spriggs & Marxer	1926		1000	38	3.8
Cryderman	1927		770	40	5.19
Lemmel	1934		3324	50	1.5
Rankin & Martin	1934		72715	111	0.016
Edwards	1939		11362	85	0.75

Thus, Maclean may be right in suggesting that roentgenograms are only efficient in diagnosing a minority of diverticula. In that event they might lead to erroneous conceptions as to the incidence of these pouches as well as to their clinical importance, particularly so, because a definite diagnosis depends exclusively on the roentgenologic findings. These are not infrequently misinterpreted, and actual diverticula are diagnosed as calcified nodes, gallstones, renal calculi, fecaliths, pancreatic calcifications, ulcer craters, redundancies, and traction pouches due to adhesions.

FIG. 9.

FIG. 10.

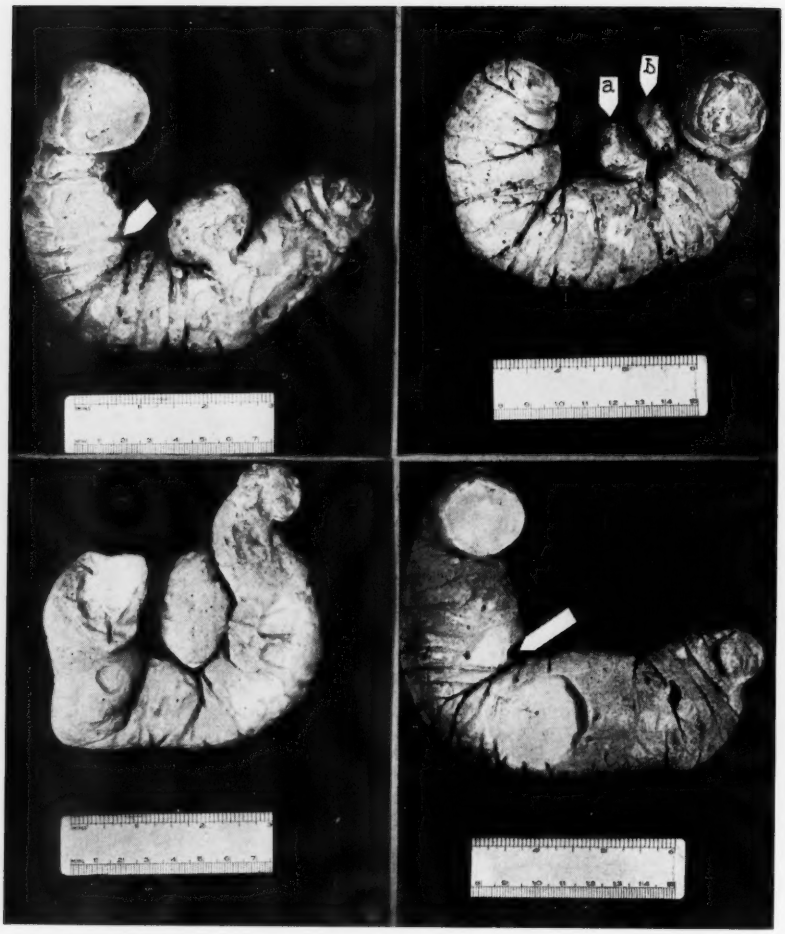


FIG. 11.

FIG. 12.

FIG. 9.—A very narrow orifice leads to a fairly large diverticulum of the third part. The arrow indicates the site of the papilla.

FIG. 10.—There are two diverticula, *a* in the third, and *b* in the fourth part of the duodenum. Both have narrow orifices.

FIG. 11.—A large oblong-shaped diverticulum of the fourth part shows a very short and narrow orifice.

FIG. 12.—The cast is of a duodenum which was composed of the first three parts only. The arrow indicates the location of the papilla.

It is quite obvious that duodenal diverticula may not always be visualized, because the barium either is expelled too rapidly or it fails to fill the pouch. Scott is of the opinion that fluoroscopic palpation is of primary importance in diagnosing diverticula, and believes that unless the barium is dammed up and manipulated into the pouch with the hands on the abdomen, little or none of the meal may enter. Heacock strongly recommends the use of a duodenal tube for filling the duodenum with barium. This is especially advisable in doubtful cases according to McKinney. Diverticula may also not always be recognized, and Costello stresses the inadequacy of the usual roentgenographic examination. He reports that in four of his six cases the diverticula were not observed until after more than one barium meal. The stomach and first part of the duodenum takes up so much of the examiner's attention, he continues, that the second, third and fourth parts of the duodenum may be neglected.

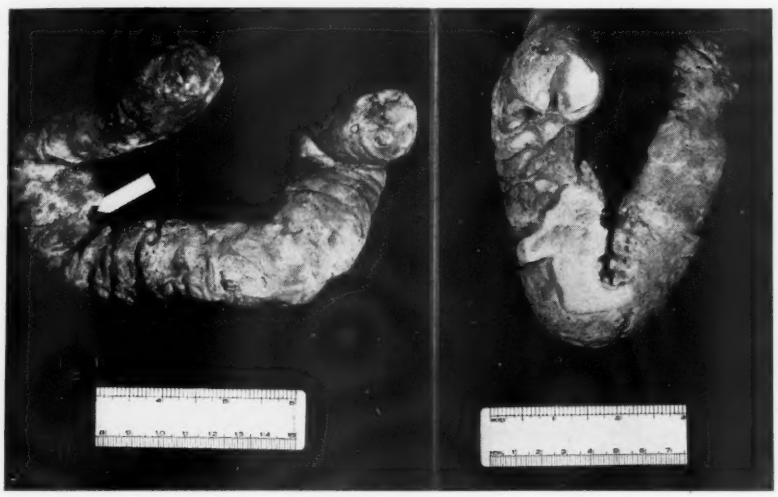


FIG. 13.—In this specimen the first and third parts are considerably larger than normal, while the second and fourth parts are quite short in proportion. Arrow indicates the location of the papilla.

FIG. 14.—Here is seen an extreme angulation at about the center of the third part, which in conjunction with the second and fourth parts give the duodenum the shape of the letter V.

Friedlander believes that serial exposures taken during screen examinations are very helpful for fixing the findings which disappear very quickly. In his opinion supine and prone positions with some rotation to the right and left and sometimes a semi-Trendelenburg position should be used, as some diverticula fill better in this way.

There may be other complicating factors as, for example, the stomach when hanging down obscures the presence of diverticula in the third part of the duodenum, or as Case points out, diverticula in the third part fail to retain barium unless the communicating orifice is small.

In spite of numerous technical difficulties encountered in diagnosing diverticula roentgenologically, a comprehensive review of the literature on the subject disclosed that these anomalies have been found quite frequently.

However, one is led to the conclusion that in most cases they have not been considered the cause of any symptoms. The validity of such an assumption can be judged from the fact that, although the condition has been known to the medical profession since 1710, when Chomel first reported such a case, no clean-cut picture has been described, by which a clinical diagnosis could be established.

There is no symptom-complex characteristic of a duodenal diverticulum, but there are a number of symptoms associated with it, and Spriggs and Marxer believe that such an association has either not been recognized, or has received but little attention, and that diverticulosis is much more common than has been hitherto recognized. Scott is of a similar opinion, and Lockwood and Maclean consider duodenal diverticula to be a clinical entity. Rankin and Martin state that in unexplained abdominal complaints diverticulosis must be thought of and ruled out.

It is evident from numerous reports that cases of such maladies most frequently are interpreted clinically as appendicitis, cholecystitis, pancreatitis, duodenitis, and gastric or duodenal ulcers, because diverticula may become inflamed or undergo other pathologic changes; they may become gangrenous and perforate or even cause obstructions, not only of the duodenum, but also of the common bile duct and the pancreatic duct as well.

The only case in which a primary carcinoma has occurred in association with a duodenal diverticulum was reported by Morrison and Feldman (1926).

Incidentally, this study of the duodenum by means of casts disclosed some of its less widely known anatomic variations, and the following observations were made:

A. The papilla of Vater in this series, as indicated by Figures 3, 7, 8, 9, 12 and 13, was found to be situated at the lower bend of the descending portion of the duodenum, at the medial side and at the posteromedial side as well (not seen in these front-view illustrations). Gray places it a little below the middle of the descending portion and at the medial side. Piersol and Cunningham locate it 3½–4 inches, and Grant 7.9 cm. beyond the pylorus. How much reliance can be placed, without reservation, upon such figures may be clearly seen when the distance from the pylorus to the papilla is compared in Figures 8 and 9.

B. That shapes, too, may vary considerably, a fact seen particularly in Figures 13 and 14.

C. That not all duodena consist of four parts; either the fourth portion is absent, as in Figure 12, or the third and fourth parts are represented by one portion ascending obliquely as in Figures 1 and 6. Some of these are angulated to such an extent that, in conjunction with the second part, they have the shape of the letter V, as in Figure 14.

CONCLUSIONS

1. This series would seem to indicate that duodenal diverticula are more common than has hitherto been believed.

2. Neither roentgenologic nor anatomic figures are conclusive regarding the actual incidence of diverticula of the duodenum.

3. It is obvious that the important points in roentgenologic diagnosis of duodenal diverticula are both a perfect technic and a genuine interest by the roengenologist in the particular subject.

4. A new method by which even the most obscure diverticula are revealed is described.

5. The papilla of Vater is not constant as to its location.

6. The duodenum is not infrequently made up of three parts instead of the classical four, and it may also vary as to its configuration and size.

The writer wishes to express his appreciation of the assistance given by Dr. J. W. Jobling, of the Department of Pathology, and particularly by Dr. D. J. Morton, of the Department of Anatomy, in connection with this paper.

He is especially indebted to Dr. Allen O. Whipple for his interest, his helpful advice and constructive suggestions, as well as for his permission to examine all bodies used in his lectures on Surgical Anatomy over a period of three years.

REFERENCES

1 Akerlund, Ake: Fortschr, a.d. Geb. d. Roentgenstrahlen, **25,** 540, 1917.
2 Alesen, L. A.: M. Rec., **149,** 261, 1939.
3 Andrews, E. W.: J. A. M. A., **77,** 1309, 1921.
4 Armstrong, J. R.: Brit. Med. Jour., **1,** 1230, 1939.
5 Avancini, von, L. P.: Deutsche Ztschr. f. Chir., **252,** 529, 1939.
6 Bainbridge, W.: Texas State M. J., **22,** 560, 1927.
7 Baldwin, W. M.: Anat. Rec., **5,** 121, 1911.
8 Basch, S.: Am. J. M. Sc., **153,** 833, 1917.
9 Bassett, V. H.: Tr. Chicago Path. Soc., **7,** 83, 1907.
10 Bauer, Theodor: Wien. klin. Wochnschr., **25,** 879, 1912.
11 Beals, J. A.: South, M. J., **30,** 218, 1937.
12 Beaver, J. L.: ANNALS OF SURGERY, **108,** 153, 1938.
13 Bell, Howard H.: Anat. Record, **21,** 229, 1921.
14 Bengolea, A. J.: Bull. et Mem. Soc. nat. de Chir., **54,** 726, 1928.
15 Boland, F. K., Jr.: Surgery, **6,** 65, 1939.
16 Buckstein, J.: Am. J. Surg., **3,** 340, 1927.
17 Buschi, Guiseppe: Virchows Arch. f. path. Anat., **206,** 121, 1911.
18 Carson, H. W.: Tr. Med. Soc. Lond., **53,** 76, 1930.
19 Case, J. T.: Am. J. Roentgenol., **3,** 314, 1916.
20 Case, J. T.: J. A. M. A., **75,** 1463, 1920.
21 Chomel, J. B. L.: Histoire de L'Academie Royale, Paris, 1710.
22 Cole, L. G., and Roberts, Dudley: Surg., Gynec. & Obst., **31,** 376, 1920.
23 Costello, C. D.: Brit. J. Radiology, **6,** 577, 1933.
24 Cronie, D.: Brit. M. J., **2,** 1021, 1933.
25 Cryderman, W. J.: Canad. Med. Assoc. J., **17,** 1455, 1927.
26 Cullen, E. K.: Arch. Surg., **2,** 542, 1921.
27 Davis, N. S.: Tr. Chicago Path. Soc., **9,** 1, 1913.
28 Downes, W. A.: ANNALS OF SURGERY, **76,** 43, 1922.
29 Edwards, H. C.: Surg., Gynec. & Obst., **60,** 946, 1935.
30 Edwards, H. C.: Diverticula and Diverticulitis of the Intestine. Bristol, John Wright & Sons, Ltd., 1939.
31 Falconer, A. W.: Lancet, **1,** 1296, 1907.
32 Finsterer, H.: Med. Klinik, **32,** 243, 1936.

33 Fischer, M. H.: J. Exper. Med., **5**, 333, 1900.
34 Fleischmann, Gottfried: Leichen Oeffnungen, **16**, 264 pp., Erlangen, 1815.
35 Fletcher, H. N., and Castleden, L. I. M.: Brit. J. Surg., **23**, 776, 1936.
36 Forssell, Gösta, and Key, Einar: Nord. med. Arkiv, **48**, 15, 1915.
37 Friedlander, G.: Brit. J. Radiol., **10**, 26, 1937.
38 Gibbon, W. H.: Radiology, **21**, 491, 1933.
39 Glickman, L. G.: Radiology, **23**, 479, 1934.
40 Grant, J. C. B.: Jour. Anat., **57**, 357, 1922–3.
41 Grant, J. C. B.: Canad. Med. Assoc. Jour., **33**, 258, 1935.
42 Habershon, S. O.: Observation on Diseases of the Alimentary Canal, 1857.
43 Hahn, O.: Klin. Wchnschr., **9**, 1922–1925, October 11, 1930.
44 Harley, G.: Tr. Path. Soc. Lond., **8**, 235, 1857.
45 Heacock, C. H.: South. Med. Jour., **22**, 348, 1929.
46 Herbst, W. P.: Minnesota Med., **10**, 364, 1927.
47 Hodenpyl, E.: Tr. New York Path. Soc., 1901.
48 Horton, B. T., and Mueller, S. C.: Arch. Surg., **26**, 1010, 1933.
49 Huddy, G. P. B.: The Lancet, **2**, 327, 1923.
50 Jacquelin, C., and Quenu, J., Jr.: Bull. et Mem. Soc. nat. de Chir., **53**, 716, 1927
51 Kaldor, J.: Annals of Surgery, **89**, 6, 1929.
52 Keith, Sir Arthur: Brit. Med. J., **1**, 376, 1910.
53 King, M. K.: South. M. J., **26**, 869, 1933.
54 Larimore, J. W., and Graham, E. A.: Surg., Gynec. & Obst., **45**, 257, 1927.
55 Letulle, Maurice: Bull. Soc. Anat. de Paris, 1898.
56 Lemmel, G.: Arch. f. Verdaungskr., **56**, 59, 1934.
57 Lewis, Dean: J. A. M. A., **76**, 783, 1921.
58 Lewis, F. T., and Thyng, F. W.: Amer. J. Anat., **7**, 505, 1908.
59 Linsmayer, Heinrich: Verhandl. d. deutsch. path. Gesellschaft, **17**, 445, 1914.
60 Lockhart, R. H.: Yale J. Biol. Med., **3**, 505, 1931.
61 Lockwood, A. L.: J. A. M. A., **98**, 961, 1932.
62 Lucinian, J. H.: Am. J. Roentgenol., **24**, 684, 1930.
63 Lund, F. B.: New England, M. J., **198**, 986, 1928.
64 MacGuire, D. P.: Intern. J. Med. & Surg., **48**, 153, 1935.
65 Mackechnie, H. N.: Annals of Surgery, **74**, 96, 1921.
66 Maclean, N. J.: Surg., Gynec. & Obst., **37**, 6, 1923.
67 Maclean, N. J.: Annals of Surgery, **85**, 73, 1927.
68 Marie, M. R.: Bull. Soc. Anat., **74**, 982, 1899.
69 McKinney, J. T.: South. Med. Jour., **24**, 753, 1931.
70 McQuay, R. W.: Annals of Surgery, **89**, 36, 1929.
71 Mendillo, H. J., and Koufman, W. B.: New England, J. Med., **219**, 432, 1938.
72 Molesworth, H. W. L.: Brit. Jour. Surg., **17**, 342, 1929.
73 Monsarrat, K. W.: Brit. Jour. Surg., **14**, 179, 1926.
74 Moore, E. C.: Arch. Surg., **1**, 513, 1920.
75 Morgagni, G. B.: DeSedibus et Causis Morborum, Naples, 1762.
76 Morrison, T. H., and Feldman, M.: Tr. Am. Gastro-Enterol. Assn., **29**, 271, 1926.
77 Morton, J. J.: Surgery, **8**, 265, 1940.
78 Nagel, H. W.: Arch. Surg., **2**, 529, 1925.
79 Nicholson, W. M.: Bull. Johns Hopkins Hosp., **56**, 305, 1935.
80 Odgers, P. N. B.: Brit. J. Surg., **17**, 592, 1930.
81 Ogilvie, R. F.: Brit. J. Surg., **28**, 362, 1941.
82 Pendergrass, R. C.: Radiology, **17**, 1216, 1931.
83 Perry, E. C., and Shaw, L. E.: Guy's Hospital Rep., **50**, 171, 1893.
84 Pilcher, L. S.: Annals of Surgery, **2**, 62, 1894.
85 Rankin, F. W., and Brown, P. W.: Surg., Gynec. & Obst., **50**, 836, 1930.

86 Rankin, F. W., and Martin, W. J., Jr.: ANNALS OF SURGERY, **100,** 1123, 1934.
87 Rich, A. R., and Duff, G. L.: Johns Hopkins Hosp., **58,** 212, 1936.
88 Ritchie, H. P., and McWhorter, G. L.: Surg., Gynec. & Obst., **25,** 485, 1917.
89 Rokitansky, Carl: Lehrbuch der pathologischen Anatomie, Vienna, 1861.
90 Rosenthal, Th.: Med. Klin., **4,** Bd. 2, 1421, 1908.
91 Roth, M.: Virchows Arch. f. path. Anat., **56,** 197, 1872.
92 Schüppel von, O.: Die Krankheiten der Gallenwege und der Pfortader. In: Handbuch der speziellen Pathologie und Therapie, **8,** 1, Halfte, 2. Abth., Div., p. 228, 1880.
93 Scott, S. G.: Brit., M. J., **1,** 346, 1931.
94 Seippel, O.: Zurich, 1895.
95 Spriggs, E. T., and Marxer, O. A.: Brit. Med. J., **1,** 130, 1926.
96 Tandler, Julius: Morphol. Jahrb., **29,** 187, Leipzig, 1902.
97 Tanner, W. E.: Lancet, **2,** 853, 1936.
98 Thorek, M.: Illinois Med. J., **55,** 64, 1929.
99 Turner, F. C.: Tr. Path. Soc. Lond., **35,** 200, 1884.
100 Turner, G. Grey: Lancet, **1,** 151, 1920.
101 Versmann, G. N.: Beitr. z. klin. Chir., **118,** 735, 1920.
102 Weiss, K.: Frankfurt, Ztschr. f. Path., 1929.
103 Wheeler, D.: Canad. Med. Assoc. J., **39,** 241, 1938.
104 Wilkie, D. P. D.: Edinburgh M. J., **11,** 219, 1913.

GASTROJEJUNOCOLIC FISTULAE: WITH SPECIAL REFERENCE TO ASSOCIATED NUTRITIONAL DEFICIENCIES AND CERTAIN SURGICAL ASPECTS

JOHN S. ATWATER, M.D.,
FELLOW IN MEDICINE, MAYO FOUNDATION
LT. HUGH R. BUTT, MC-V (S) U.S.N.R.,*
AND
JAMES T. PRIESTLEY, M.D.,
DIVISION OF SURGERY, MAYO CLINIC
ROCHESTER, MINN.

FEW DISEASE SYNDROMES are capable of producing such a diversity of nutritional deficiencies so rapidly as the syndrome which may accompany gastrocolic or gastrojejunocolic fistula. For many years the deficient nutritional status of patients who have this condition has been one of the largest contributing factors to the high mortality rate attendant on the surgical treatment of this lesion. In the past, at least part of the high surgical mortality rate in such cases could be attributed to inadequate medical treatment, both preoperatively and postoperatively. But in view of the present knowledge of nutrition, these inadequacies should be overcome and the added surgical risk entailed by nutritional deficiencies should be largely eliminated.

A series of patients who had gastrojejunocolic fistula was studied with a twofold purpose: First, for review of the clinical observations and determination of the incidence, predisposing factors, signs, symptoms and the ultimate outcome of patients who have gastrocolic fistulae; second, for consideration of the clinical evidence of associated metabolic deficiency diseases. In focusing attention on the poor nutritional status of these patients, we hope that the need for adequate preoperative and postoperative care will be emphasized in the achievement of the best surgical results.

The material presented herein was selected from the records of the Mayo Clinic in the seven-year period from January, 1935, to January, 1942. Forty-two patients who had surgically-proved gastrocolic or gastrojejunocolic fistulae were encountered during this period. In 40 of these 42 cases the fistulae were associated with benign disease, and in the other two, complicated malignant disease of the upper portion of the gastro-intestinal tract was associated. Certain of these cases were included in a series reported previously by Gray and Sharpe.[1]

Gastrocolic or gastrojejunocolic fistulae may be the sequelae of three different types of disease process: First, there are those which complicate cancer of the stomach or colon; second, those which complicate gastrojejunal ulcers; and third, those associated with miscellaneous diseases, such as abscess in the peritoneal cavity, visceral tuberculosis, syphilis, trauma, and

* On leave from Division of Medicine, Mayo Clinic.

possibly congenital deformities[2] of the gastro-intestinal tract. Those which complicate gastrojejunal ulcers are, by far, the most common.

HISTORICAL ASPECTS

Gastrocolic Fistula and Cancer.—Haller,[3] in 1755, made the first report of a case in which gastrocolic fistula complicated a malignant process of the stomach. Zweig[4] had collected 70 cases of this type by the turn of this century, and, in 1940, Thomas[2] reported or mentioned a total of 141 cases. Since in this group gastrocolic fistula generally occurs only after the malignant disease is far advanced, and since modern diagnostic methods make possible the earlier recognition of the underlying cancer in most cases, fewer and fewer cases of this type have been reported.

Postgastrojejunal Ulcer Group.—Wolfler[5] performed the first anterior type of gastro-enterostomy in 1881, and von Hacker[6] performed the first posterior type of gastro-enterostomy in 1885. Four years later (in 1889), Braun[7] made the first report of a gastrojejunal ulcer, and, in 1903, Czerny[8] made the first report of a case in which gastrojejunocolic fistula followed gastrojejunal ulcer. By 1940, Thomas had collected references to 207 cases in which gastrocolic or gastrojejunocolic fistula had followed gastrojejunal ulcer.

INCIDENCE

Thus, until the advent of gastric surgery for the treatment of duodenal ulcer, most gastrocolic fistulae were associated with cancer of the stomach and colon. As gastro-enterostomy began to be performed frequently, the incidence of gastrocolic and gastrojejunocolic fistula increased proportionately to the number of gastrojejunal ulcers which developed. Probably because of anatomic reasons, fistulae of this type seldom occur after the performance of anterior gastro-enterostomy. It is also of interest to notice that after posterior gastro-enterostomy has been performed for gastric ulcer, gastrojejunocolic fistula develops infrequently, as do gastrojejunal ulcers.

Reports of the incidence of gastrojejunal ulcer which occurs after gastro-enterostomy vary widely. Lahey[9] stated that an incidence of 15 per cent ascribed to the condition would be fairly accurate. However, in various reports, such as those by Walton,[10] from the London Hospital, Allen,[11] from the Massachusetts General Hospital, and Walters and Clagett,[12] from the Mayo Clinic, agreed that the incidence is about three to four per cent. In arriving at such an incidence, the latter authors assumed that in the several series, on which they based their conclusions, the operation of gastro-enterostomy had been properly selected for the individual patient and correctly performed.

The incidence of gastrojejunal ulcer occurring after gastric resection, when that operation has been considered properly the operation of choice for a given patient with duodenal ulcer, and when it has been performed in an approved manner, is lower than it is after gastro-enterostomy. Likewise, the incidence of gastrojejunocolic fistula after gastric resection is lower

under these circumstances. The incidence of gastrojejunocolic fistula in the presence of gastrojejunal ulcer has been reported to be from 11 to 14 per cent. Walters and Clagett reported this incidence to be 13.6 per cent. Allen has reported it to be 14 per cent.

FACTORS OF AGE AND SEX

Lahey and Swinton[13] stated that gastrojejunocolic fistulae usually occur among patients from 30 to 45 years old. In Rife's[14] series the average age of the patients was 46 years. In Walters and Clagett's series the age group was from 40 to 60 years. In the present series the average age of the patients at the clinical onset of gastrocolic or gastrojejunocolic fistula, excluding two cases in which the condition was secondary to malignant disease, was approximately 45 years. The two patients who also had cancer were 65 and 72 years old, respectively. Of the remaining 40 patients, the youngest was 28 years old and the oldest was 68 years old. From this point onward, only the 40 patients whose background condition was benign will be considered herein, and the two patients just mentioned, in whom fistulae developed secondary to carcinoma, will be omitted.

It is difficult to state the incidence of duodenal ulcer among men and women. However, Robertson and Hargis[15] in a pathologico-anatomic survey of duodenal ulcers found evidence of healed or active ulcers in 11.8 per cent of cases. Their observations were based on results of necropsy as performed 2,000 times. Certain clinical roentgenologic studies support their conclusion, for in one 12-month period at the Mayo Clinic duodenal ulcers were found in 12 per cent of the 16,000 gastroduodenal examinations performed during that period.[16] Balfour and Eusterman stated that peptic ulcer occurs four times more often among men than among women; that gastrojejunal ulcer occurs seven times more often among men than among women, and that gastrojejunocolic fistula almost never occurs among women.

In this series proper, there were no cases in which gastrocolic or gastrojejunocolic fistula had occurred among women. In all the 40 cases the fistulae had developed among men as sequelae to gastrojejunal ulcers. The two patients who had fistulae which complicated a malignant process were women.

FACTOR OF PREVIOUS OPERATIONS

In all instances the fistulae followed some surgical procedure on the upper part of the gastro-intestinal tract. In 23 cases (58 per cent) the fistulae followed posterior gastro-enterostomy; in 14 cases (35 per cent) the fistulae followed an unidentified type of gastro-enterostomy. Gastrectomy preceded one instance of fistula, and an unidentified type of operation preceded still another instance. In no case was a fistula preceded by performance of an anterior type of gastro-enterostomy. This brief summary brings the total number of cases to 39.

In the fortieth case there had been a previous surgically-proved gastro-

jejunocolic fistula. The patient in this case was of especial interest. After 15 years of typical symptoms and roentgenologic evidence of a duodenal ulcer, the ulcer perforated. The perforation was closed and a posterior type of gastro-enterostomy was performed. Four years later symptoms of a gastrojejunal ulcer were noticed. This ulcer, in turn, after another interim of six years, gave rise to a gastrojejunocolic fistula. At operation, the posterior gastro-enteric anastomosis was disestablished, the fistula was excised, and the openings in the jejunum and stomach were closed. That portion of the colon containing the fistulous opening was exteriorized, as is done in performance of a Mikulicz operation. Soon thereafter the old duodenal ulcer became reactivated and perforated acutely. Simple closure of the perforated ulcer was performed. The ulcer remained active, symptoms of pyloric obstruction developed and, at the end of eight months, a posterior type of gastro-enterostomy was performed because there was such a large inflammatory mass surrounding the duodenal ulcer that gastric resection could not be accomplished. After this operation the patient complained of unmistakable symptoms of gastrojejunal ulcer and a gastrojejunocolic fistula soon developed for the second time. Operation was again performed and the fistula was excised, but the patient succumbed on the sixth postoperative day, from pneumonia.

INTERIM BETWEEN INITIAL OPERATION AND ONSET OF SYMPTOMS OF GASTROJEJUNOCOLIC FISTULA

It is interesting to observe the interval that elapsed between the initial operative procedure and the onset of symptoms attributable to gastrojejunocolic fistula. In 17 cases a fistula developed within a year after operation; in 24 cases a fistula developed within two years after operation; in 27 cases a fistula had developed by the third year after operation; in 28 cases a fistula had developed by the fifth year after operation; and in 35* cases a fistula had developed by the end of the tenth year after operation. In the remaining five cases a fistula occurred between 10 and 20 years after the original operations (Table I).

TABLE I

INTERIM BETWEEN OPERATION AND ONSET OF SYMPTOMS IN 40 PATIENTS WHO HAD GASTROJEJUNOCOLIC FISTULAE

Years Since Operation	No. of Cases in Each Period	Cumulative No. of Cases in Each Period
0 to 5	28	28
6 to 10	7	35
11 to 15	2	37
10 to 20	3	40

SIGNS AND SYMPTOMS

Diarrhea.—Diarrhea was present in 39 of the 40 cases. Often diarrhea was the presenting complaint, and it was of a varying degree of severity. In 27 cases diarrhea was intermittent in character or moderately severe—

* Each number, of course, includes the total in the group which immediately precedes it.

meaning that 6 to 12 stools were passed daily. In the remaining cases passage of stools occurred more frequently.

The stools were described as being either "loose and watery" or "mushy and infantile" in form and consistency. They were often copious and the color usually was described as "light yellow" or "gray-white." In some instances they were described as "foamy," "greasy" and "able to float on water." It is noteworthy that the stools rarely were bloody. In 28 per cent of cases the stools contained abnormal quantities of undigested food particles. In certain cases undigested food from the patient's previous meal was recognized in the stools within from two to four hours after eating. Thirty-five per cent of the patients had stools which contained an abnormal amount of fat. In a few instances determination was made of the percentage of fat in the stools, and in each case an increased quantity of fat was reported. Approximately a fourth of the patients complained of the necessity of the nocturnal passage of multiple stools.

Some effort was made to correlate the size of the ostia of the fistulous tract with the severity of the diarrhea. In general, those patients in whom the diameter of the ostia was less than two centimeters had more intermittent and milder diarrhea than those patients in whom the diameter of the ostia was larger than two centimeters.

Loss of Weight.—In general, loss of weight was directly proportional to several factors; namely, duration of the disease, severity of the diarrhea and, in turn, the relative diameter of the ostia of the fistulous tract. An occasional patient exhibited no change in weight, but others lost as much as 50 pounds (23 Kg.) within five or six months after the development of the gastrojejunocolic fistula.

Loss of Strength.—Loss of strength was severe in a few cases, moderate in most cases, and absent in only five per cent of cases (Table II). Seventy-eight per cent of the patients had moderately severe loss of strength. Dehydration was frequent and paralleled the severity of the diarrhea and the loss of strength and weight.

TABLE II

GENERAL AND MISCELLANEOUS DEFICIENCIES OBSERVED IN 40 PATIENTS WHO HAD GASTROJEJUNOCOLIC FISTULAE

	Number of Observations
Emaciation and dehydration	23
Nutritional edema	10
Ascites	1
Loss of strength	38
Loss of weight	31
Disease of liver	
Value for serum protein reduced*	15
Fatty liver (observed at operation or at postmortem)	7
Dye retention (Grade 2)	1
Definitely low albumin-globulin ratio	3
Low normals	6
Salt craving	1
Loss of body hair	1

*Eighty-eight per cent of patients tested had definitely lowered values for protein in the serum; lowest value was 3.7 Gm., and average value was 4.9 Gm. per 100 cc.

Stercoraceous Eructation and Vomiting.—Another prominent complaint was belching or vomiting of material which the patient said tasted like, and which often appeared to be, fecal substance. Approximately half of the patients complained of fecal belching, 38 per cent complained of fecal vomiting and 30 per cent gave no history of either. There were two cases in which vomiting of material not fecal in character occurred. In many instances the test meal produced material similar to feces in odor and color. In view of the nature of the gastric contents, values for hydrochloric acid as noted in analysis of a test meal were thought to be of little significance.

EVIDENCE OF NUTRITIONAL DEFICIENCIES

Gastrojejunocolic fistula causes a disease syndrome in which there is a unique pathologic anatomic situation especially conducive to the development of nutritional deficiencies. Other diseases, such as chronic ulcerative colitis, sprue, celiac disease and the chronic diarrheas, are capable of producing similar states of nutritional deficiency. In the case of gastrojejunocolic fistula, however, the mechanical factor, rather than any disease process, is foremost. When gastrojejunocolic fistula is present, food taken orally actually spends little time in the gastro-intestinal tract. It is shunted out of the stomach and into that portion of the bowel in which it is known that only little absorption of food takes place. In fact, the total time which orally ingested food spends in the entire gastro-intestinal tract may be as short as from one to two hours. Fluids, also, have but little time for absorption by the gastro-intestinal tract, and as a result most stools are loose and liquid.

In the presence of gastrojejunocolic fistula, in addition to the rapid passage of gastric contents through the alimentary tract, as just described, there is a decrease in the available absorptive intestinal surface with which food comes into contact. Some nutriment is obtained, but most of the food and fluid passes on largely unchanged, except by mastication. This is recognized readily by the presence of abnormally high quantities of food fats and undigested food particles in the watery stools. The disturbance in water balance is manifest by the prominent dehydration which most patients display.

When gastrojejunocolic fistula has occurred there is ample clinical evidence of inadequate provision of the important foodstuffs, namely carbohydrates, proteins, fats and vitamins. Undernutrition generally is outstanding. Mention has been made of the striking losses in weight and strength encountered. Twenty-three patients (58 per cent) exhibited a marked degree of emaciation and dehydration (Table II).

It is well known that deprivation of proteins provokes hypoproteinemia and that this, in turn, may induce changes productive of shifts in body fluids. Edema interpreted as being "nutritional" in origin was present among ten patients (25 per cent) and one of these patients had associated ascites which could be explained on no other basis. The value for proteins in the serum was estimated among a number of patients, and in almost every instance it was found to be low. Values for protein in the serum varied

from the low figure of 3.7 to 4.9 Gm. per 100 cc. In some cases values for both albumin and globulin were lowered. Tests of hepatic function were not performed regularly; however, in seven cases the surgeon at the time of operation or the pathologist at the time of necropsy was impressed sufficiently to remark on the fatty condition of the liver.

Deficiency of iron in the body will produce a hypochromic form of anemia; a deficiency of Castle's antianemic principle will produce a macrocytic form of anemia. Such was found to be true in this study. Examination of the blood revealed definite changes in many cases. The lowest erythrocyte count was 3,363,000, and the lowest value for hemoglobin was 8.4 Gm. per 100 cc. In 68 per cent of cases the erythrocyte count was less than 4,500,000 and in 15 per cent of cases it was less than 4,000,000. In 52 per cent of cases the value for hemoglobin was below the normal range. Examination of blood smears disclosed macrocytosis to be present in 14 per cent of cases.

When the manifold evidence of general undernutrition was considered; that is, loss of weight and of strength, dehydration and emaciation, peripheral edema, ascites, hypoproteinemia and anemia, it seemed probable that certain vitamin deficiencies should find clinical expression. Such was the case. These patients had been observed during a period in which refined laboratory measurements of vitamin deficiency diseases were being developed. With few exceptions, evidence of avitaminosis had to be adduced from the clinical observations alone. The guideposts for the clinical recognition of vitamin deficiency states were many. A solitary deficiency state seldom existed. The number of instances of vitamin deficiency observed was in direct proportion to the severity of the diarrhea and the duration of the disease.

Patients who have chronic diarrhea are known to have a greater requirement for vitamins than normal persons have.[17] The decrease in the power of adaptation to dark is one of the earliest manifestations of vitamin A deficiency. Two patients displayed a marked degree of this, as nyctalopia or "night blindness" (Table III). At the time of observation the test for adaptation to dark was not used clinically as widely as it is now, so that the exact degree of dysadaptation to dark was not observed.

Absence of members of the vitamin B-complex provoked the most striking evidence of vitamin deficiency states. A third of the vitamin deficiencies observed were in this category. Lack of the thermolabile antineuritic component, thiamine, was disclosed by the presence of burning, superficial parasthesias, and in one instance severe peripheral neuritis was noted. The patient who complained of the latter condition also had a typical type of niacin deficiency, pellagra. The cutaneous lesions were characteristic. Glossitis was observed twice, and was ascribed to a lack of either niacin or riboflavin. Closely related was the occurrence of cheilosis in three patients; in one of them a noninfectious type of conjunctivitis was noted. These were considered to be typical instances of B-complex deficiency, and in the light of present knowledge could be attributed to a deficiency of riboflavin.

TABLE III

TYPES OF VITAMIN DEFICIENCY OBSERVED AND SYMPTOMS THEREOF IN 40 PATIENTS WHO HAD GASTROJEJUNOCOLIC FISTULAE

Deficiency and Symptoms of	No. Having Symptoms
Vitamin A:	
Nyctalopia	2
Thiamine:	
Paresthesias	1
Peripheral neuritis	1
Riboflavin:	
Cheilosis	3
Glossitis	2
Conjunctivitis	1
Niacin:	
Pellagra	1
Ascorbic acid:	
Lowered values in blood*	11
Ecchymoses	1
Vitamin D:	
Tetany (carpopedal spasm)	1
Vitamin K:	
Prolonged Quick prothrombin time	6
Total	30

*These 11 patients were all that had been tested for this acid. In all 11 the values were lower than normal.

Both clinical and laboratory observations were used to detect vitamin C deficiency. Eleven patients had been tested for values for ascorbic acid in the blood. All had definitely lowered values for this acid. One patient in this group had multiple ecchymoses.

Quick prothrombin times had been determined in 14 cases, and in this group a definitely prolonged time was present in 43 per cent. One patient had a Quick prothrombin value of 90 seconds as compared with a normal time of 20 seconds. This case was one of particular interest, being one in which the condition was most severe, the patient displaying pellagrous lesions, carpopedal spasm and other evidences of nutritional disturbances.

Preoperative Preparation.—Inasmuch as a large proportion of patients who have gastrojejunocolic fistula exhibit clinical evidence of marked deficiency disease, it is obvious that a most energetic effort to restore the patient to proper physical and chemical health prior to operation should be considered of utmost importance. Such an effort can be made by means of good preoperative care. Treatment of this type also should be continued postoperatively. There is no specified textbook procedure. The primary objective is to restore the patient to good nutritional balance as quickly and as economically as possible. Gray and Sharpe reported a mortality rate of 61.5 per cent in a series of cases in which operation had been performed for gastrojejunocolic fistula without adequate preoperative preparation of the patients, in contrast with a mortality rate of 27.7 per cent in a group of patients who received adequate preoperative care.

Adequate quantities of fluids in the form of physiologic solution of sodium chloride and solutions of glucose should be given by hypodermoclysis. Transfused blood and plasma are of value. In addition to the fact that they con-

tribute a certain amount of fluid, the former are valuable in that they tend to restore protein to the plasma. The use of extracts of iron and liver may be indicated. A high caloric, high vitamin and low residue diet is suggested.

No elaborate plan of dosage of vitamins is necessary so long as the amounts administered are in excess of the optimal standards as adopted by the National Research Council[18] for an active person. Preparation should be carried on not for one day preoperatively, but should extend over a longer period. In general, the preoperative program should be planned so as to replenish generously each of the sorely depleted nutritional stores of the body, and as a rule requires one week to two weeks.

SURGICAL ASPECTS

Many different surgical procedures have been suggested for the treatment of gastrojejunocolic fistula. As experience with the surgical management of this lesion has increased, the relative appreciation of the advantages and disadvantages of the various procedures has become more nearly accurate. Some surgeons have endeavored to accomplish all that is necessary in a single-stage procedure, whereas others have employed operations of several stages in the hope of reducing the operative risk.

In general, the various plans of surgical procedures can be divided into three main categories: First, a preliminary procedure of some type performed in the hope of reducing the risk associated with subsequent removal of the fistula; second, removal of the fistula without the making of any appreciable effort to prevent further formation of an ulcer or ulcers; third, removal of the fistula in association with performance of partial gastrectomy for the purpose of correcting the immediate condition and also preventing subsequent gastric, duodenal or jejunal ulceration.

Preliminary jejunostomy or colostomy are examples of the first type of procedure. In former years jejunostomy occasionally was employed as a procedure by which it was hoped that the general nutritive state of the patient might be improved prior to performance of the more extensive procedure of removing the fistula. This procedure is only partially successful as a rule, and for several reasons it is not employed now as often as in the past. With the current availability of various vitamin preparations and nutritive supplements for parenteral use, as much usually can be accomplished by the use of such preparations in combination with transfusions of blood and plasma as by the insertion of a jejunostomy tube for feeding purposes. More recently, Pfeiffer and Kent[19] have suggested preliminary colostomy, and in our experience this has been most helpful. Usually, a portion of the transverse colon between the hepatic flexure and the site of the gastrojejunocolic fistula is exteriorized for the purpose of drainage. The improvement which takes place in almost every patient after this procedure is most striking. Generally, there is a definite gain in weight associated with improvement of appetite, amelioration of anemia and lessening of the diarrhea. After performance of colostomy proximal to the fistulous tract, regurgitation of colonic contents

into the stomach and thereby into the small intestine is prevented. Presumably, such regurgitation is at least in part responsible for certain important alterations in the patient's general condition. It appears obvious that fecal material which is constantly retained in the stomach would cause a certain amount of inflammatory reaction in that organ. In addition, much of this fecal material might well leave the stomach through the duodenum and thereby be absorbed to some degree in the small intestine, an abnormal state of affairs which might readily result in deleterious effects. Furthermore, preliminary colostomy has an additional advantage of distinct value in that it eliminates all fecal material from the region of the fistula and permits thorough cleansing of the stomach, jejunum and colon in the vicinity of the fistula prior to performance of the corrective type of surgical procedure. In addition, after closure of the fistula in the colon no fecal material remains in this portion of the bowel, a fact which should improve the chance for good healing. It appears that this factor in itself adds greatly to the safety of removal of the fistula.

Operations directed mainly toward removal of the fistula without performance of an associated procedure of significance to prevent further ulceration are not employed as frequently today as they were formerly. We refer to surgical disestablishment of the gastro-enteric anastomosis, with excision of the fistula and closure of the openings in the stomach, jejunum and colon, associated or unassociated with some type of local operation on the pylorus and duodenum. Although procedures of this type do correct the immediate situation, they offer little protection against the possibility of recurrent ulceration. Usually, procedures of this type have been employed for the patient who is in exceedingly poor condition and for whom it seems advisable to keep the surgical procedure to an absolute minimum. In the past jejunostomy often has been performed in association with this form of operation. With current methods of preoperative preparation, however, jejunostomy does not often seem necessary.

The corrective operation of choice, if the condition of the patient permits it (and generally it will, if proper preliminary attention, both medical and surgical, has been carried out), consists of surgical disconnection of the gastro-enteric anastomosis, excision of the fistula, closure of the openings in the jejunum and colon, and generous resection of the stomach, preferably with the establishment of a posterior Pólya type of gastrojejunal anastomosis. The stomach is joined to the jejunum distal to the site in the jejunum at which the previous gastro-enteric stoma existed. This form of surgical treatment not only eliminates the fistula but also offers definite protection against recurrent ulceration. As a rule, such a procedure can be accomplished without undue risk, provided, as said before, adequate preliminary preparation has been carried out. In our opinion, therefore, the treatment of choice for gastrojejunocolic fistula consists in proper preliminary preparation with the administration of fluids, vitamins and the transfusion of blood if necessary, followed by performance of colostomy proximal to the fistula. After a

period of from three to four months, if the patient is in poor general condition, the fistula is then excised and the stomach is resected. Subsequently, when the patient's condition permits it, the colonic stoma is closed. Employment of a regimen of this type will be followed, we think, by more and more satisfactory results.

Results.—Results of the surgical treatment of gastrojejunocolic fistula in the past have not been satisfactory; however, it seems reasonable to believe that currently more favorable results are being obtained. If the one patient in whom a recurrent gastrojejunocolic fistula formed is considered as representative of two cases in which operation was performed for this condition, it results that there were 11 postoperative deaths in a group of 41 cases (Table IV). This means that the mortality rate was

TABLE IV

RESULTS OF SURGICAL TREATMENT: 41 CASES OF GASTROJEJUNOCOLIC FISTULAE AMONG 40 PATIENTS

	Gastric Resection		
Result	Performed, 17 Cases	Not Performed, 24 Cases	Total, 41 Cases
Operative deaths	2	9	11
No further record	10	3	13
Recurrent distress from ulcer	1	9	10
Good health	3	3	6
Died, unrelated causes	1	0	1
Died, massive hematemesis*	0	1	1
Total	17	25	42

*This patient is also listed as having had recurrent ulcer distress; hence the totals of 25 and 42, respectively in the final two columns.

approximately 27 per cent. There are several factors which seem most important in the reduction of the operative mortality rate. These include a full appreciation of the nutritional deficiencies which this lesion may cause and adequate preoperative preparation to compensate for these abnormalities. In addition, the performance of colostomy, carried out proximal to the fistula, as a preliminary procedure prior to removal of the fistula and gastric resection, reduces the risk of the latter procedure. Postmortem examination, which was performed on ten of the 11 patients who succumbed after operation, revealed that peritonitis and bronchopneumonia were the two complications which contributed most frequently to the fatal outcome.

Of the 30 patients who survived operation, 13 were not heard from after their original dismissal, and consequently the condition of their health is unknown. Of the remaining 17 patients, six were reported to be in good health and ten had had further distress of the ulcerous type, to the time of this report. In this regard it is of interest to consider the result obtained in relation to the type of operation performed. In the entire 41 cases,* gastric resection was performed seventeen times and in the remaining 24

* Counting, as said previously, the one patient who twice underwent operation for gastrojejunocolic fistula as representative of two cases. There were, of course, 40 patients in the series.

cases an operation which did not include gastric resection was carried out. Of the ten patients who experienced further distress of the ulcerous type after operation, only one had undergone gastric resection. In contrast, three of the six patients who were reported to be "cured" had undergone gastric resection. These results substantiate the merit of gastric resection in the presence of gastrojejunocolic fistula.

SUMMARY

Gastrojejunocolic fistula results in a unique mechanical gastro-intestinal shunt. By its mechanism a decrease occurs in the available absorptive intestinal surface. The presence of such a pathologic anatomic pattern is conducive to the development of nutritional disturbances.

In the present series, with two exceptions, gastrojejunocolic fistula in all cases followed gastrojejunal ulcer, the ulcer having been a sequela to a previous operation, usually gastro-enterostomy. In 42 per cent of the 40 cases the fistula occurred within a year after the original operation. In one patient a gastrojejunocolic fistula developed for the second time. All patients were males. The average age of the patient at the clinical onset of the condition was 45 years. The estimated incidence of gastrojejunocolic fistula in the presence of gastrojejunal ulcer varies between 11 and 14 per cent.

Diarrhea and fecal belching or vomiting are the most common symptoms of this condition. Evidence of malnutrition is manifold. Extreme loss of weight and strength and the presence of dehydration, emaciation, hypoproteinemia, nutritional edema, anemia and multiple vitamin deficiency states are characteristic. Such vitamin deficiencies as night blindness, peripheral neuritis, paresthesias, pellagra, glossitis, conjunctivitis, cheilosis, ecchymoses with decreased values for ascorbic acid in the blood and hypoprothrombinemia have been observed. The primary object of preoperative care should be to replenish each of the depleted body nutritional stores, and in so doing, to restore the patient to satisfactory chemical and physical health, so that surgical operation can be performed with the least possible risk.

The surgical treatment of choice for gastrojejunocolic fistula consists of resection of the stomach after the stomach, jejunum and colon have been detached from their fistulous connection. In certain cases preliminary colostomy may be advisable. In the present series of 41 cases (40 patients, of whom one underwent operation twice for fistula) the operative mortality rate was approximately 27 per cent. There are reasons for the belief that this mortality rate will be materially reduced in the future. Results after gastric resection are definitely superior to those which follow mere removal of the fistulous tract in a procedure unassociated with partial gastrectomy.

REFERENCES

[1] Gray, H. K., and Sharpe, W. S.: Preoperative Management of Gastrojejunocolic Fistula. Arch. Surg., **43**, 850–857, November, 1941.

[2] Thomas, J. R.: Gastrocolic and Gastrojejunocolic Fistulae: Report of Five Cases. Mil. Surgeon, **87**, 232–243, September, 1940.

[3] Haller: Quoted by Walters and Clagett.[12]
[4] Zweig: Quoted by Walters and Clagett.[12]
[5] Wolfler: Quoted by Thomas.[2]
[6] von Hacker; Quoted by Thomas.[2]
[7] Braun: Quoted by Walters and Clagett.[12]
[8] Czerny: Quoted by Walters and Clagett.[12]
[9] Lahey, F. H.: Diagnosis and Management of Gastrojejunal Ulcer and Gastrojejunocolic Fistula. Surg. Clin. North America, **20,** 767–780, June, 1940.
[10] Walton, A. J.: Gastrojejunal Ulceration. Brit. J. Surg., **22,** 33–55, July, 1934.
[11] Allen, A. W.: An Aseptic Technic Applicable to Gastrojejunocolic Fistula. Surgery, **1,** 338–348, March, 1937.
[12] Walters, Waltman, and Clagett, O. T.: Gastrojejunocolic Ulcer and Fistula. Am. J. Surg., **46,** 94–102, October, 1939.
[13] Lahey, F. H., and Swinton, N. W.: Gastrojejunal Ulcer and Gastrojejunocolic Fistula. Surg., Gynec. & Obst., **61,** 599–612, November, 1935.
[14] Rife, C. S.: Gastrojejunocolic Fistula. Am. J. Surg., **40,** 73–88, April, 1938.
[15] Robertson, H. E., and Hargis, E. H.: Duodenal Ulcer: An Anatomic Study. Med. Clin. North America, **8,** 1065–1092, January, 1925.
[16] Eusterman, G. B., and Balfour, D. C.: The Stomach and Duodenum. Philadelphia, W. B. Saunders Company, 1935, 958 pp.
[17] Spies, T. D., and Butt, H. R.: Vitamins and Avitaminosis. In Duncan, G. G.: Diseases of Metabolism. Philadelphia, W. B. Saunders Company, chap. 8, 1942, pp. 366–502.
[18] Committee on Food and Nutrition, National Research Council: Recommended Allowances for the Various Dietary Essentials. J. Am. Dietet. A., **17,** 565–567, June–July, 1941.
[19] Pfeiffer, D. B., and Kent, E. M.: The Value of Preliminary Colostomy in the Correction of Gastrojejunocolic Fistula. ANNALS OF SURGERY, **110,** 659–666, October, 1939.

ACUTE ILEUS*

ANALYSIS OF 130 CASES OPERATED UPON AT THE PRESBYTERIAN HOSPITAL, NEW YORK CITY, FROM 1936 TO 1939, INCLUSIVE: WITH USE OF MILLER-ABBOTT TUBE IN 1938 AND 1939

BEVERLY CHEW SMITH, M.D., AND FREDERICK T. VAN BEUREN, M.D.
NEW YORK, N. Y.

THIS REPORT completes a 24-year study of acute ileus at the Presbyterian Hospital by the same authors. The entire study has been divided into six four-year periods. The mortality by periods is recorded in Table I:

TABLE I

MORTALITY BY PERIODS

Period	Number of Cases	Late Cases†	Mortality
1916–1919 inclusive	60	81%	66.6%
1920–1923 inclusive	80	66%	53.7%
1924–1927 inclusive	108	61%	44.4%
1928–1931 inclusive	72	71%	48.6%
1932–1935 inclusive	130	60%	28.4%
1935–1939 inclusive	130	68%	23.8%

Toward the end of this period the following observations were outstanding:

1. The Staff has become, so to speak, more conscious of the possibility of intestinal obstruction in cases of obscure abdominal complaint.

2. The status of enterostomy has been reviewed, indications for the operation clarified, and its employment decreased.

3. Dehydration has been recognized, methods developed to determine the degree present, and various properly prepard solutions have been made easily available for its early correction.

4. Electrolyte deficiency has been chemically studied and early replacements made.

5. Roentgenograms have become a dependable diagnostic modality.

6. Gastric suction has effectively supplemented or replaced lavage, bringing with it chemical electrolytic problems requiring laboratory solution.

7. Intestinal intubation with the Miller-Abbott tube has largely supplemented or replaced gastric suction.

8. Sulfonamide drugs in the peritoneal cavity, parenterally and *per os,* have become accepted therapy.

9. The parenteral peristaltic drugs combined with rectal treatments have been more frequently used.

10. Spinal anesthesia has become, both as to drug and technic, safer and more effective.

---

* Read before the New York Surgical Society, February 24, 1943.

† Signs and symptoms of obstruction present on admission for 48 hours or more.

11. The percentage of late cases admitted to the Hospital has remained about the same during 24 years, namely, 60 to 69 per cent.

We still believe it advisable that more than one observer engage in such a review. Controversial points of diagnosis were freely discussed before recording.

This 1935–1940 series contains 130 cases which, by chance, is exactly the same as the 1932–1935 group.

This report is not to be confused with that of Leigh, Nelson and Swenson[2] from the same institution in 1940, concerning the use of the Miller-Abbott tube as an adjunct to surgery of small intestinal obstructions. In their series many cases were diagnosed but recovered with conservative therapy without operation. Their classification was: I. Noninflammatory, (A) paralytic and (B) mechanical obstruction; II. obstruction with peritonitis; and III, obstruction with gangrene, whereas we have continued our original classification of complete, incomplete obstruction, and paralytic ileus. *All* cases in our series were operated upon. This classification, because of the Miller-Abbott tube, will require subsequent modification in order to include nonoperative cases, which have been diagnosed roentgenologically.

For comparison, a table of the present and previous series is presented (Table II):

TABLE II

COMPARISON OF PRESENT AND PREVIOUS SERIES[1]

	Total Number	Late Cases	Recovered	Died	Mortality
(1932–1935, Inclusive)					
Series	130	60%	94	36	27.6%
Complete obstruction	91	63%	78	13	14.3%
Incomplete obstruction	26	88%	15	11	42.3%
Paralytic ileus	13	69%	1	12	92.3%
(1936–1939, Inclusive)					
Series	130	68%	99	31	23.8%
Complete obstruction	103	63%	84	19	18.4%
Incomplete obstruction	16	88%	14	2	12.5%
Paralytic ileus	11	90%	1	10	90.9%

A comparison of these series shows eight per cent more late cases in 1936–1939 group. This should be accompanied, on past experiences, with a higher group mortality, but this is not so. The group mortality was 3.8 per cent less. The predominating factors in these results were the use of the Miller-Abbott tube, continuous gastric suction, and a greater correction of dehydration. Sulfonamide drugs were not used in the last series. The greatest difference in group mortality lies in the incomplete cases, where there was a decrease from 42.3 to 12.5 per cent. This we believe due primarily to the use of the Miller-Abbott tube. The incomplete group reduction was the greatest factor in reducing this series mortality.

TABLE III

ANALYSIS OF CASES GROUPED AS TO ETIOLOGY

1. ACUTE COMPLETE MECHANICAL OBSTRUCTION

(*Series 1935—1939, Inclusive*)

	Number of Cases	Recovered	Died	Mortality
Old postoperative adhesions or bands (2 with gangrene)	47	41	6	12.7%
Recent adhesions or bands (recent peritonitis)	14	12	2	14.3%
Carcinoma of small intestine and colon	8	5	3	37.5%
Strangulated hernia (8 with gangrene)	28	20	8	28.0%
Volvulus of intestine	5	5	0	0
Intussusception	1	1	0	0
Totals	103	84	19	18.4%

The mortality of this group in the 1932–1935 series was 14.3 per cent, due to a mortality in the old adhesions group of 2.5 per cent in 41 cases. This illustrates that the cases of one series may, by chance, be more desperate than in another comparable one.

TABLE IV

ACUTE INCOMPLETE MECHANICAL OBSTRUCTION

(*Series 1935—1939, Inclusive*)

	Number of Cases	Recovered	Died	Mortality
Old postoperative adhesions	11	9	2	10%
Recent adhesions (recent peritonitis)	4	4	0	0%
Neoplasm of intestine	1	0	1	100
Strangulated hernia	1	1	0	0 %
Totals	16	13	2	12.5—

In the previous series this group contained 26 cases, with a mortality of 42.3 per cent. The reduced mortality in this group was the result of fewer colon carcinoma cases and the use of the Miller-Abbott tube.

TABLE V

PARALYTIC ILEUS

(*Series 1935—1939, Inclusive*)

	Number of Cases	Recovered	Died	Mortality
Appendicitis, with acute diffuse peritonitis	4	1	3	75%
Mesenteric thrombosis, with peritonitis	3		3	100%
Acute diffuse peritonitis (cause undetermined)	3		3	100%
Acute diffuse peritonitis following pelvic operation	1		1	100%
Totals	11	1	10	90.9%

We define paralytic ileus as those cases with a generalized intestinal distention, without mechanical obstruction, with diffuse peritonitis or other profound pathology, and believe they should not be operated upon except to remove a focus.

The paralytic ileus group mortality of 90.9 per cent indicates its gravity prior to the use of sulfonamide drugs. The mortality in 13 paralytic ileus cases in the previous series was 92.3 per cent. Thus, in eight years, combining these two series, the mortality in 24 cases of paralytic ileus is 91.6 per cent.

TABLE VI

ANESTHESIA

(*Series 1935—1939, Inclusive*)

	Number of Cases	Recovered	Died	Mortality
Local	14	11	3	21.4%
General	67	52	15	23.8%
Spinal	49	39	10	21.2%
Total	130			

Anesthetics should be thoughtfully adapted to each case. Local was employed for the sickest, and the least surgery. Spinal—a single or continuous injection—became the anesthetic of choice. It collapsed intestine, which gave better exposure, ease of closure, and shortened operative time. Disadvantages were lowered blood pressure, anoxemia, and shock, which were combated by ephedrine, intravenous fluids and oxygen.

In complete obstructions fluid replacement averaged 2,900 cc. daily during their acute illness, which was 700 cc. more than the previous series. Daily intake rose for short periods to 5,000 cc. or more, depending upon other pathology.

In the same group, peristaltic drugs were administered in 59 cases, with a mortality of 13.5 per cent. In the previous series they were used in 60 cases, with a mortality of 15 per cent. In the 39 cases they were not used, the mortality was 18 per cent. The Miller-Abbott tube, with rectal treatments postoperatively, decreased their use. In the *early* cases in which gastric and Miller-Abbott tubes were not used they were more effective.

Diagnostic roentgenograms taken with patients sitting up, lying on right and left sides, were positive for gas and fluid levels in 94 per cent of 83 cases operated upon, and inconclusive or negative in six per cent. Those with negative roentgenologic diagnosis, with obstruction, were such early cases that fluid levels were not present, and distension alone was not sufficient to warrant a positive diagnosis. A negative roentgenogram should not delay operation if the history and physical findings are definitive.

The negative roentgenograms were on incomplete obstructions which were not severely distended. None of these died. This would suggest that a negative roentgenogram may indicate a good prognosis. In the presence of a negative film, and clinical evidence of obstruction, exposures should be frequently repeated if the patient is not operated upon.

TABLE VII

ENTEROSTOMY FOR COMPLETE OBSTRUCTION

	Primary	Mortality	Secondary	Mortality
1932—1935, incl.	30	23.3%	12	23%
1936—1939, incl.	25	16%	5	60%

The result of combining the last two series of enterostomy cases is tabulated in Table VIII:

TABLE VIII

AGGREGATE STATISTICS OF MORTALITY FOLLOWING ENTEROSTOMY IN THE LAST TWO SERIES

(*Series 1932—1935 and 1936—1939, Inclusive*)

	No. Cases	Mortality
Enterostomy for complete obstruction	72	27.7%
Enterostomy for incomplte obstruction	18	59%
Enterostomy for paralytic ileus	18	88.8%

Operative Treatment.—In all series, this varied from division of a single band, to colon or small bowel resection for advanced pathology. Enterostomy has been less frequently performed since the advent of the Miller-Abbott tube, particularly in the secondary group, and, recently, less in the primary group. We prefer the Witzel technic, without passing the tube through omentum or suturing intestine to the peritoneum.

The Miller-Abbott tube was first used in The Presbyterian Hospital in December, 1937. Its use has steadily increased, until now, for medical and surgical conditions, it has been employed in approximately 700 cases. We employ a graduate nurse to supervise these tubes and, except for records concerning each case, she has no other duties.

TECHNIC OF EMPLOYMENT OF THE MILLER-ABBOTT TUBE

The tube is used as follows: The nasal passage is inspected for adequate patency; together with the pharynx it is cocainized; the balloon is folded over the tip, well lubricated and passed through the nostril into the stomach. Sipping water aids its more rapid passage. The tip is placed at the pylorus

TABLE IX

SERIES OF 130 CASES—MILLER-ABBOTT TUBE EMPLOYED IN 36, OR 27.6 PER CENT

(*Series 1935—1939, Inclusive*)

Diagnosis	No. Cases	Died	Mortality
Paralytic ileus	5	4	80%
Mechanical complete and incomplete obstruction	31	3	9.6%
Totals	36	7	19.1%

TABLE X

COMPLETE AND INCOMPLETE OBSTRUCTION

(*Series 1935—1939, Inclusive*)

	No. Cases	Died	Mortality
Miller-Abbott tube	31	3	9.6%
Gastric suction	40	10	25%
No tube or suction	48	8	16.6%
Totals	119	21	17.6%

TABLE XI

PARALYTIC ILEUS

	No. Cases*	Died	Mortality
Miller-Abbott tube	5	4	80%
Gastric suction	6	6	100%
Totals	11	10	90.9%
* Sulfonamide drugs and Miller-Abbott tube	1	0	0

TABLE XII

TOTAL SERIES

INCLUDING MECHANICAL COMPLETE, INCOMPLETE AND PARALYTIC CASES

(Series 1935—1939, Inclusive)

	No. Cases	Died	Mortality
Miller-Abbott tube	36	7	19.1%
Gastric suction	46	16	34.7%
Without Miller-Abbott tube or gastric suction	48	8	16.6%
Totals	130	31	23.8%

under the fluoroscope with the tube lying along the greater curvature. It is held lightly by adhesive against the nose (not cheek) for if the slack lay along the greater curvature there is sufficient tubing in the stomach to allow the tip to reach the third portion of the duodenum without introducing more, which may coil or kink.

The following observations concerning the Miller-Abbott tube warrant emphasis:

The progress of the tube can be noted by fluoroscopy and recorded by roentgenograms. Sufficient roentgenray can be administered under such circumstances to affect the skin. Roentgenologic Department observations of the tube should be timed on the chart.

Its rapid passage through the pylorus is facilitated by manipulation under the fluoroscope. This requires experienced observations in a minimum of time. Suction deflates the stomach. Fluids are allowed in order to propel the tube to the pylorus. The patient preferably, but not necessarily, lies on his right side. Once through the pylorus the balloon is inflated with 20cc. of air and allowed to progress of its own accord. Peristalsis carries the inflated balloon downward, causing a noticeable tug. In paralytic ileus the tube may not advance so rapidly, and requires more attention to deflate atonic coils of intestine. In the absence of peristalsis it may be advanced by doctor or patient, but not faster than six inches every two hours, in order to prevent coiling in the stomach. It is frequently irrigated with 40–60 cc. of saline to insure its patency, dilute intestinal contents and deflate successive loops. Patients are so grateful for the relief after gastric deflation that they often cooperate with subsequent tube care. The position of the tube tip can be determined by (1) aspiration of bile or jejunal content; (2) the time required for fluid by mouth to be aspirated; and (3) by lack of resistance to inflation of the balloon if it remains in the stomach. We do not hesitate to transport sick cases to fluoroscopy for manipulation of the tube if it has not passed out of the stomach in 6–12 hours. During this time continuous gastric suction is emptying the stomach.

The time, amount and character of the aspirated fluid should be charted, together with hematocrit, plasma specific gravity, plasma protein and blood chloride determinations. Chloride replacement should be approximately five grams for each liter aspirated. Continuous aspiration with the tube tip in the jejunum returns one-half to two-thirds of the fluid taken by mouth,

whereas about one-third returns with the tip in the ileum. Frequent fluoroscopic or roentgenologic examinations determine the site and rate of progress of the tube.

Often, regurgitated loop-content requires withdrawal of the tube tip into that loop, aspirating it, and again allowing the tube to advance. The stomach may become distended while the tube tip is deflating the jejunum, in which case a gastric lavage by a Levin tube may be necessary. The tube should be left *in situ* until (1) the obstruction is relieved or passed; (2) it has reached the cecum; or (3) the character of the aspirated fluid becomes normal intestinal contents, and the Roentgenologic Department agrees that its diagnostic possibilities have been exhausted.

The tube was left *in situ* for an average of five and one-half days. The longest time was 39 days. The average fluid recovered was 3,300 cc. for the ordinary case. The largest amount was 36,000 cc. in 27 days.

At operation, it has served as a guide to the site of obstruction.

Successful intubation deflates, improves blood supply and neromuscular mechanism of peristalsis, permits absorption and correction of fluid and electrolyte imbalance, diagnosis of site and extent of local pathology, and converts a toxic, distended late case into either a comparative early or a nontoxic, nonobstructed one.

With the tube *in situ*, high protein fluids can be given and hypoproteinemia be corrected by other than plasma and whole blood. A tube which has coiled into a knot may remain patent but may not pass a partial obstruction. During intubation, clinical signs and symptoms of compromised blood supply of intestine must be constantly kept in mind and watched for. They are often difficult to ascertain and may occur in the absence of fever, leukocytosis and tenderness. The development of gangrene of the intestine while an ileus toxemia is being combatted, constitutes the greatest danger associated with the Miller-Abbott tube. Fortunately, if patients are closely watched, this occurs infrequently.

The tube may be retarded by a kink, band, foreign body, tumor, inflammation of the wall or extrinsic pressure. Following deflation, barium may be injected through the tube and films may reveal the nature of the local pathology. The fact that the barium advances but the tube does not, confirms the degree of the mechanical obstruction. Multiple obstructions may be demonstrated. A site of cryptic bleeding may be localized by gross appearance or chemical tests upon aspirated fluid.

The following adverse incidents have occurred in our experiences with this tube:

Patients may not tolerate it. Irrational patients may remove it, and restraint increases their restlessness. It is difficult, almost impossible, to get a Miller-Abbott tube to pass through a gastrojejunal stoma. It may be coughed or vomited up. It is poorly tolerated in the presence of pneumonia. Nasal pathology makes it locally uncomfortable. The nasopharyngeal portion

should be cleansed of dried secretions, oiled and replaced several times a day. Esophagitis has been reported, but it has not manifested itself clinically in our cases. Laryngitis has not been a serious complication. The tube may not pass through the pylorus because of gastric atony, distension or local pathology. It has been regurgitated from the duodenum. Inflation of the balloon prevents this. It has coiled and knotted in the stomach and intestine. The balloon has been broken by overinflation and the injection of fluid. The orifices for fluid and air should be plainly marked. The tip may become lodged in a partial obstruction and convert it into a complete one. An inflated balloon may act similarly. The small intestine has intussuscepted itself on the tube. It has delayed operation and permitted gangrene of the intestine to occur during the period of treatment. Incorrect interpretation of the patient's response to treatment, and delay in making these observations, may unnecessarily delay operation. Large bowel treatments (lavage, *etc.*) administered when the tube is in the terminal ileum or cecum have been recovered through the oral end of the tube. Fluids given for nourishment, particularly those containing milk, may block the tube and necessitate its withdrawal for cleansing.

Confronted with an early case of obvious intestinal obstruction, without severe distension, confirmed roentgenologically, and without clinically evident dehydration, one must decide whether emergency operation or Miller-Abbott tube intubation should be undertaken on the merits of the particular case. Given a late case, similarly proven, distended, and dehydrated, the Miller-Abbott tube should seriously be considered for preoperative treatment, for its greatest usefulness has been observed in this group. If signs and symptoms of intestinal gangrene are suspected or are evident, operative delay because of Miller-Abbott tube treatments will produce a fatality.

For resections of obstructed as well as unobstructed right colon carcinoma cases, this tube has largely replaced enterostomy as a decompressing procedure and has permitted more successful one-stage operations.

DISCUSSION

This last group (1935–1939) of a 24-year series showed the lowest mortality, we believe, due to an earlier diagnosis, aided largely by roentgenology, adequate fluid and electrolyte replacement, better anesthesia, the use of peristaltic drugs and rectal treatments, and, particularly, the employment of the Miller-Abbott tube.

The Miller-Abbott tube and gastric suction were not employed in early cases with simple pathology who had an easy, short operation, whose intestine showed peristalsis, and who responded to drugs and rectal treatments.

Small intestinal roentgenologic studies should be made upon cases of obstruction which have recovered without operation. Findings may indicate an interval exploration.

Early diagnosis of obstruction remains difficult. Roentgenologic examina-

tion has been the single, outstanding, dependable diagnostic aid. Diagnosis should be made before the roentgenologic findings become positive, but this is often difficult. Obstruction occurring during another illness is even more difficult to diagnose.

The same type of errors of diagnosis and treatment seem to recur in each series, but are least outstanding in the latest series.

Delay in certain instances is unwarranted in attempting to make a new treatment work. We have seen this, in turn, with hypertonic saline, peristaltic drugs and the Miller-Abbott tube.

Prolonged operations upon seriously ill, dehydrated, distended patients is just as dangerous today as it was a quarter of a century ago.

Injudicious choice of anesthesia may be fatal. It should be adapted to the patient.

A scar of a previous abdominal operation remains an indication of a possible obstruction in an abdominal complaint.

We are still impressed by the number of obstructions that follow abdominal gynecologic operations.

The positive roentgenogram may not follow the suspected clinical signs of obstruction for some time, but it may be positive earlier than the clinical diagnosis. When the roentgenologic diagnosis is not positive, the entire situation should be reviewed by a senior observer.

As important as electrolytic balance is, it can be further upset by overtreatment.

Witzel enterostomy technic has demonstrated the following as compared to that of Kader:

(a) Fewer have had to be closed operatively.

(b) They have functioned better and less has escaped around the tube.

(c) With removal of the tube the intestinal opening has leaked less and closed more quickly.

Enterostomy as an adjunct to the treatment of intestinal obstruction should not be discarded. It has been more effective as a primary than secondary procedure.

Principles concerning viable and nonviable intestine should be reviewed before closing an abdomen.

Chart records could be more easily classified if there was, in the written operative procedure, a concise note as to the duration of preoperative symptoms, the presence of collapsed and distended intestine, an exact description of the condition of the intestine, and the obstructing mechanism. The last, at times may be most difficult.

Aerobic and anaerobic cultures should be taken of peritoneal fluids and resection sites at the completion of the operation.

Six to eight grams of sulfanilamide should be left in the abdomen in cases with an odoriferous fluid or in which the intestine has been opened. It is not a good substitute for poor surgery.

Too many instances of secondary peritonitis have followed contamination from hernia sacs. Better protection should be practiced.

Richter herniae are dangerous because of difficulty of diagnosis; even the roentgenogram misses them, and the segment of intestine involved is often nonviable.

Methods of preventing peritoneal adhesions, including the size and position of the incision through the abdominal wall, are still an important part of the ritual of any abdominal operation.

REFERENCES

[1] van Beuren, F. T., Jr., and Smith, Beverly.: Acute Ileus. ANNALS OF SURGERY, 107, 321, 1938.

[2] Leigh, O. C., Jr., Nelson, John H., and Swenson, Paul C.: The Miller-Abbott Tube as an Adjunct to Surgery of Small Intestinal Obstructions. ANNALS OF SURGERY, 111, 186, 1940.

ISLET CELL CARCINOMA OF PANCREAS, WITH METASTASIS

HAROLD A. HANNO, M.D., AND ROLAND W. BANKS, M.D.

PHILADELPHIA, PA.

FROM THE DEPARTMENTS OF PATHOLOGY, GASTRO-ENTEROLOGY, AND SURGERY, GRADUATE HOSPITAL, UNIVERSITY OF PENNSYLVANIA, PHILADELPHIA, PA.

A NUMBER of islet cell tumors of the pancreas, both benign and malignant, have been recorded in the literature. Nicholls,[45] in 1902, reported the first unequivocal islet tumor, an adenoma discovered as an incidental finding at autopsy. Fabozzi,[18] in 1903, described five cases, four of which showed involvement of other organs. During the next few years additional cases of islet cell adenoma were found.[29, 28, 44, 12] Vecchio,[53] in 1914, described an islet cell tumor found in aberrant pancreatic tissue. Lang[39] was the first to report the finding of multiple adenomata of the pancreas. Reviewing the subject in 1926, Warren[54] could find only 16 reports of islet cell tumors, none of which involved distant or neighboring organs, and he added four cases of his own. In none of the cases which he reported was a clinical summary included. He ventured the opinion that these lesions "probably never give rise to trouble during life and have no clinical significance."

However, following the discovery of insulin by Banting and Best,[2] in 1922, and the recognition of the effects of insulin overdosage,[22, 3] the tumors of the islet cells of the pancreas soon were resurrected from the limbo of mere pathologic curiosities. Interest in these tumors mounted because of the clinical picture which some of them were found to present.

Credit belongs to Harris[26] for suggesting, in 1924, the possibility of hypersecretion of insulin as the causative factor in certain cases of spontaneous hypoglycemia. This hypothesis was confirmed when, in 1927, Wilder, Allan, Power, and Robertson[61] reported their case of the physician who clinically presented a typical picture of severe hypoglycemia and who showed at operation and at autopsy an islet cell tumor of the tail of the pancreas, with metastases to the liver and the regional lymph nodes. Assay of the tumor tissue revealed the presence of a markedly increased insulin content. Warren[55] regarded the demonstration of insulin in the tumor metastases in this case as laying the foundation for the clinical entity of hyperinsulinism and he considered it as the final proof of the etiologic relationship of insulin lack to diabetes mellitus. Since the report of Wilder and his associates, the presence of insulin in increased amounts in the tumor tissue in cases of islet cell neoplasm, with hypoglycemia, has been demonstrated several times.[14, 7, 30]

The incidence of tumors of the islands of Langerhans is a matter of some question. Whipple and Frantz,[59] in 1935, reported only 61 cases, only one of which showed metastases. Three years later Whipple[57] listed 74 cases of islet cell tumor, with hypoglycemia, and, in 1940, Frantz[21]

was able to gather 96 cases. In 1941, Keating and Wilder[37] reported 106 cases. This number has been increased to 134 in Whipple's[58] recent article. More recently a number of additional cases have been recorded.[38, 47, 48, 42, 63] Autopsy estimates as to the frequency of islet cell tumors vary from one per 10,000[40] to five in 4,010 consecutive necropsies.[46] It is of interest to note, in passing, that an islet cell adenoma has been found in a mouse[31] and that malignant neoplasms, with metastases, have been described in dogs on two occasions.[50, 8]

Islet cell adenomata are of interest because of the clinical hypoglycemia which often occurs. Campbell, Graham and Robinson[10] state that in only 20 per cent of the cases of islet cell tumor does hypoglycemia exist. In the series of Whipple and Frantz,[59] however, 31 tumors unassociated with clinical hypoglycemia were found at autopsy as opposed to 30 such tumors accompanied by hypoglycemia; and in Duff's[16] group of 90 case reports there were 64 hypoglycemic cases. The absence of thorough clinical records may well account for at least a portion of the number of cases of islet cell tumor reported with no mention of hypoglycemia.

Undeniable islet cell carcinoma with involvement of neighboring structures or metastases to distant organs is rare. A number of cases of islet cell tumors involving only the pancreas have been thought to be malignant on histologic examination.[30, 19, 52, 43, 13, 35, 4, 53] Frantz[21] lists 21 cases in her total of 91 islet cell neoplasms limited to the pancreas. These tumors have been variously labeled malignant, questionably malignant, adenocarcinoma Grade 1, and of low grade malignancy, because, microscopically, they showed a number of the characteristics associated commonly with malignancy. Lack of capsulation, incomplete encapsulation, blood vessel invasion, and cellular anaplasia with frequent mitotic figures have been described in these tumors. However, the 15 patients in Frantz's group of tumors of questionable or low-grade malignancy who survived surgery have shown no metastases or recurrences in follow-ups of from eight weeks to ten years, only four cases having follow-ups of less than a year. According to Duff,[16] the patient of Howland, Campbell, Maltby, and Robinson,[30] who was operated upon in 1929 for a "malignant" islet cell tumor limited to the pancreas, was reported well 12 years following surgery and 19 years after the onset of his symptoms of hypoglycemia. Frantz[21] expressed doubt as to whether all of the tumors in this "locally malignant" group constitute real malignancies, although she admitted that metastases might occur at later dates. Duff,[16] and Brunschwig[9] concur in the belief that a histologic appearance of malignancy in cases of islet cell tumor is not a valid index of malignancy. This point deserves emphasis. The present authors feel that, as far as can be determined at present, the only true criterion of malignancy in islet cell neoplasms is the presence of involvement of other organs by direct extention or by metastasis, the microscopic characteristics of the tumor tissue notwithstanding. Further reports of longer follow-ups in questionable cases are, of course, to be desired.

A careful search of the literature to date (December, 1942) has revealed only 21 cases of islet cell tumor with involvement of other organs, to which group we should like to report another. These cases, with the available pertinent data, are tabulated in Table I. Willis'[62] unusual case of widespread malignancy may possibly represent an instance of islet cell carcinoma. We are omitting the case reported by Berardinelli[6] because this is, in all probability, a malignancy of the common acinar type. Ballinger's[1] report of a widespread malignancy in which the pancreas was not involved is not included in the tabulation because his surmise that the primary tumor arose from ectopic pancreatic tissue in the liver may not be valid.

It is of interest to note that Fabozzi,[18] in describing his five cases of islet cell tumor, apparently did not realize that he was dealing with an unusual type of neoplasm, for the conclusion which he drew from his observations was that malignant tumors of the pancreas nearly always arise from the cells of the islands of Langerhans.

Case Report.—B. G., white, male, age 68, was admitted, September 10, 1942, to the Graduate Hospital on the Gastro-Intestinal Service of Dr. H. L. Bockus. His chief complaint was upper abdominal pain. He dated the onset of his illness to 21 months ago, at which time he struck himself with a wrench and broke several ribs. After two weeks in bed he had no complaints until 19 months before admission when he experienced a severe, sharp pain in the left lower chest. This was accompanied by dyspnea, fever, and chills, but there was no cough or hemoptysis. A diagnosis of pleurisy was made. It was at this time that he first noted frequent cold sweats accompanied by marked weakness which would come on at about 6 A.M. Though never unconscious, on one occasion he could not speak. His wife would give him milk and bread and he would feel much better. Because of his pleurisy he was treated by bed rest for nine months, during which period he had marked swelling of both legs.

The upper abdominal pain began about nine months prior to admission. It was dull and aching in character, not very well localized, and involved first the right side and then both sides of the upper abdomen, but for the most part was epigastric in location. Though the pain was worse when he was hungry, food or soda gave no noticeable relief, but belching seemed to alleviate the discomfort. The pain was aggravated by deep inspiration. He had lost about 30 pounds in weight during the year prior to hospitalization, and associated with this was loss of strength and energy. He complained of exertional dyspnea, but gave no history of vomiting, jaundice, clay-colored or tarry stools, or a change in bowel habit. Since their onset nine months previously, the episodes of morning weakness, relieved by food, have recurred frequently.

Physical Examination.—This revealed a rather florid-faced, elderly white male who showed evidence of marked weight loss. He was in no acute distress and the sensorium was clear. The scalp, eyes, ears, and nose were essentially negative. There was partial edentia, and the tongue was rather red and showed very slight atrophy of the markings at the margins. The lungs were moderately emphysematous. The heart size was within normal limits, and the rate, rhythm, and sounds were normal. B. P. 142/80. Examination of the abdomen revealed a large, hard, irregularly rounded, dome-shaped mass in the epigastrium. The mass was tender and moved very slightly with deep respiration. The liver edge was palpable three centimeters below the right costal border, and the area of splenic dullness was enlarged. Rectal examination disclosed a moderately enlarged prostate, which was thought to be benign. Bilateral hydrocele was noted, and there was slight pitting edema of the right ankle. Neurologic examination was negative.

TABLE I

A TABULATION OF CASES OF ISLET CELL CARCINOMA, WITH INVOLVEMENT OF OTHER ORGANS

Case No.	Author	Year	Age	Sex	Survival After Onset of Symptoms	Hypoglycemia	Site in Pancreas	Size of Tumor	Involvement of Other Organs
1.	Fabozzi[18]	1903	65	?	No clinical data	No clinical data	Head	Orange-sized	Gallbladder
2.	Fabozzi[18]	1903	30	M.	No clinical data	No clinical data	Head	Not mentioned	Liver, and hilum of spleen
3.	Fabozzi[18]	1903	56	M.	No clinical data	No clinical data	Tail	"Head of fetus"	Stomach, liver, and retroperitoneal nodes
4.	Fabozzi[18]	1903	?	?	No clinical data	No clinical data	Head	Not mentioned	Gallbladder
5.	Zanetti[65]	1927	56	M.	No clinical data	No clinical data	Entire pancreas	Not mentioned	Stomach, liver, peritoneum, and regional nodes
6.	Wilder, *et al.*[61]	1927	40	M.	21 months	Present	Tail	Not mentioned	Liver, perigastric and peripancreatic nodes
7.	Lloyd[40]	1929	?	?	No clinical data	No clinical data	Not mentioned	Not mentioned	Stomach
8.	Hamdi[25]	1932	52	M.	No clinical data	No clinical data	Tail	2 x 2.5 cm.	Liver
9.	Judd, Faust, and Dixon[36]	1934	18	F.	4 months	Present	Not mentioned	Not mentioned	Liver (no autopsy)‡
10.	Jacobsen[33]	1934	36	M.	1 to 2 years	Present	Head	Grapefruit-sized	Liver
11.	Bickel, Mozer, and Junet[7]	1935	56	M.	10 mos. (approx.)	Present	Body and tail	7 x 2.5 x 5 cm.	Skin, pleura, peritoneum, adrenals, heart, lungs, liver, pleura, and gallbladder
12.	Evangilisti[17]	1935	65	M	No clinical data	No clinical data	Body and tail	Not mentioned	Omentum, and liver
13.	Cragg, Power, and Lindem[14]	1937	41	F.	7 months	Present	Entire pancreas	Not mentioned	Liver, pancreatic, mesenteric and aortic nodes
14.	Joachim and Banowitch[32]	1938	31	F.	3 months	Present	Distal half	"Tangerine-orange"	Regional nodes (no autopsy)‡
15.	Seckel[49]*	1939	36–38	M.	1 year	Present	Proximal half	4 x 2 x 2 cm.	Liver, one adrenal, spine, lungs, pleura, peripancreatic, periaortic and mediastinal nodes
16.	Duff[15]	1939	32	M.	3 months	Absent	Entire pancreas	Not mentioned	Regional nodes, liver, peritoneum, pericardium, thyroid, adrenals, kidney, and vertebrae
17.	Duff[15]	1939	60	M.	7 months	Absent	Entire pancreas	Not mentioned	Liver, kidney, lung, myocardium, skin, abdominal and mediastinal nodes
18.	Duff[15]	1939	45	M.	5½ months	Absent	Entire pancreas	Not mentioned	Abdominal, mediastinal, cervical and axillary nodes, liver, stomach, gallbladder, adrenal, peritoneum, pituitary, ureter, kidney, bladder, lungs, and brain
19.	Flinn, *et al.*[20]	1941	45	F.	6 mos. (approx.)	Present	Head	Not mentioned	Liver, and retroperitoneal nodes
20.	Gray[24]†	1942	48	F.	4½ years	Present	Tail	1.8 x 1.8 x 1.3 cm.	Liver
21.	Quarrier and Bingham[47]	1942	73	M.	3½ weeks	Present	Tail	3 cm. in diameter	Liver, and regional nodes
22.	Hanno and Banks	1943	68	M.	19 months	Present	Tail	8 x 8 x 6.5 cm.	Liver

*This case has been reported also by Brunschwig[9], Gomori[23], and Cannon.[11]

†This case has been reported also by Joslin *et al.*[34]

‡Operative findings.

Note: All but two cases were autopsied.

Laboratory Data.—Erythrocytes, 5.1 millions; hematocrit, 39; hemoglobin 13 grams (78%); leukocytes 6700, with 72% neutrophiles, 7% monocytes, 1% basophiles, and 20% lymphocytes; urinalysis was negative except for traces of albumin and occasional leukocytes; flocculation and complement fixation tests for syphilis were negative; serum bilirubin was less than 0.2 mg. per cent; prothrombin time by Quick's method 13 seconds (100%); serum phosphatase 5.2 and 4.4 Bodansky units; blood urea nitrogen 17 mg. per cent; cholesterol 216 mg. per cent; fasting blood sugar 54 mg. per cent, serum albumin 3.85 grams per cent; serum globulin 2.79 grams per cent. There was seven per cent dye retention 30 minutes after the intravenous injection of two milligrams of bromsulfalein per kilogram of body weight. The stools were repeatedly negative for occult blood and positive for bile. Gastric analysis revealed 48 units of free acid and 74 units of total acid at the end of one hour. Roentgenologic study of the colon was negative and a barium progress meal disclosed only some displacement of the stomach to the left.

The patient was placed on a smooth meat-free diet and given a maintenance dose of one and one-half grains of digitalis daily. At 1:30 A.M. the third morning after admission, he began to moan and perspire profusely, complaining also of weakness. He was unconscious by the time he was seen by a physician. A blood sugar was taken and 25 cc. of 50 per cent glucose was administered intravenously, whereupon the patient "awoke" quickly, sat up in bed, and drank some orange juice. The blood showed 44 mg. per cent of sugar.

At 5:00 A.M. the following morning a similar episode occurred. Six minims of adrenalin subcutaneously was without effect, but 15 cc. of 50 per cent glucose revived him. The next morning the blood sugar at the time of another attack was 21 mg. per cent. That same day, at 11:00 A.M., he had another period of unconsciousness which was quickly controlled by intravenous glucose.

Because of the repeated attacks of hypoglycemia, he was placed on a high fat, high protein diet with a small midnight feeding. No further episodes of hypoglycemia occurred while he was on this regimen.

After an eight-hour fast a glucose tolerance test was performed, using 100 grams of glucose dissolved in water. The values obtained are shown below. At the beginning of the test the patient complained of weakness and was perspiring. Again at the end of five hours he began to perspire profusely and complained of weakness. The test was not carried out for the full six-hour period because between the fifth and sixth hours the patient became unconscious and had mild generalized convulsions, which promptly responded to intravenous glucose.

Fasting—	34 mg. per cent of sugar
½ hour —	73 mg. per cent of sugar
1 hour —	104 mg. per cent of sugar
2 hours—	151 mg. per cent of sugar
3 hours—	133 mg. per cent of sugar
4 hours—	69 mg. per cent of sugar
5 hours—	32 mg. per cent of sugar
5 to 6 hours—	26 mg. per cent of sugar

Operation.—September 23, 1942: A celiotomy was performed by Dr. Walter Estell Lee. The liver was found to be enlarged and studded with circumscribed whitish-gray masses which varied in size from that of a pea to that of a walnut. A large mass posterior to the peritoneum, and just inferior and adherent to the spleen, was palpated. The exact origin of the mass could not be definitely determined. A biopsy of one of the liver nodules was taken, and the abdomen closed.

Pathologic Examination.—Dr. Eugene A. Case. "The specimen received is a small biopsy of a tumor nodule in the liver. Very little liver tissue is present, its place being taken by an epithelial tumor whose cells have a well-stained, rounded

nucleus with scattered granules of chromatin and a considerable clear cytoplasm. Some of the cells are elongated and some rounded or polyhedral, and they are arranged in nests or processes suggesting glandular origin. Mitosis occurs but is not frequent. The fibrous stroma is rather loose in texture and fairly abundant. From the clinical history it is highly probable that this tumor arose from cells of the islands of Langerhans, though we were unable to demonstrate the granules found in these cells. *Pathologic Diagnosis:* Metastatic carcinoma of islet cell origin."

The patient's postoperative course was rapidly downhill. Glucose intravenously either every four hours as a 50 per cent solution or continuously as a 10 per cent infusion was necessary to prevent severe hypoglycemia. He developed dyspnea and tachycardia and went into shock several times during the second postoperative day, being revived each time by intravenous glucose. That evening he went into shock once more and expired a few moments later. Postmortem examination was performed by one of us (H. A. H.) 16 hours after death.

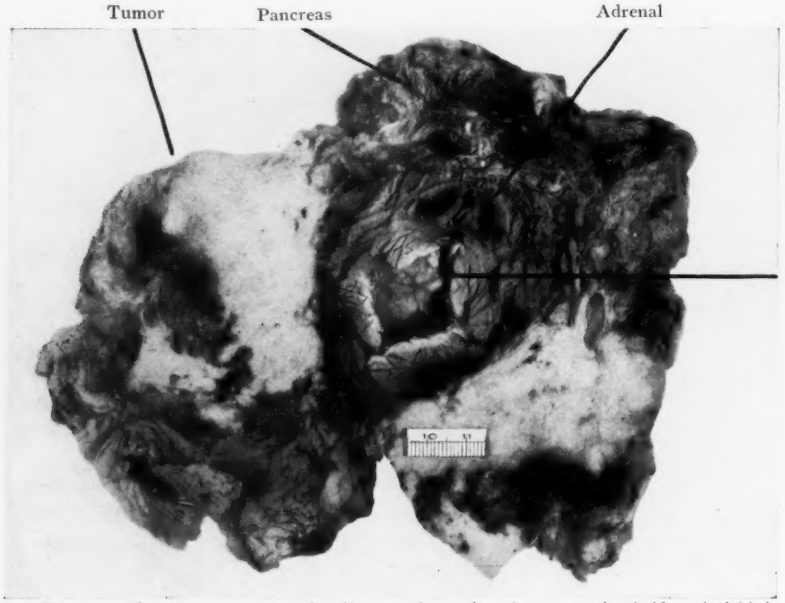


FIG. 1.—The tumor mass and adjacent tissue has been cut in half and folded back. The distal half of the pancreas can be seen to be continuous with the tumor mass proper, and the hemisected left adrenal is visible in the fatty tissue between the halves of the tumor.

Necropsy Findings.—Gross: Recent midline abdominal surgical wound; bilateral avascular pleural adhesions; healed calcified tuberculosis of the left lung; pulmonary congestion; coronary sclerosis; atherosclerosis of the aorta; retention cysts of the kidneys; possible benign nephrosclerosis; benign prostatic hyperplasia; bilateral hydroceles; bilateral cysts of the hydatids of Morgagni; carcinoma of the tail of the pancreas; and widespread metastatic carcinoma of the liver.

When the abdomen was opened the stomach was found to be deviated to the left, and in the left upper quadrant was a large firm mass the size of a small grapefruit. The spleen was firmly adherent to the upper and anterior surfaces of the mass and the splenic flexure of the colon was adherent to it anteriorly and inferiorly. The omentum was adherent to the tumor mass anteriorly, and posteriorly the left adrenal was attached to the mass by a thickness of soft fatty tissue. The left kidney was freely movable with respect to the tumor and adherent structures and was not involved by the neoplasm. Posteriorly, the pancreas was found to be directly continuous with

the tumor mass; there was no line of cleavage between the mass proper and the distal portion of the pancreas. The head and the body of the pancreas were grossly normal; the tumor involved the tail.

The tumor mass was roughly rounded, but presented a number of nodular projections. On the whole, it was firm and whitish, but several soft areas were noted which, on section, were found to contain a sticky, purplish material. The cut surface of the tumor revealed areas of focal hemorrhage and presented a fasciculated appearance. The mass (Fig. 1) measured 8 x 8 x 6.5 cm., and the combined weight of the pancreas and tumor was 370 grams.

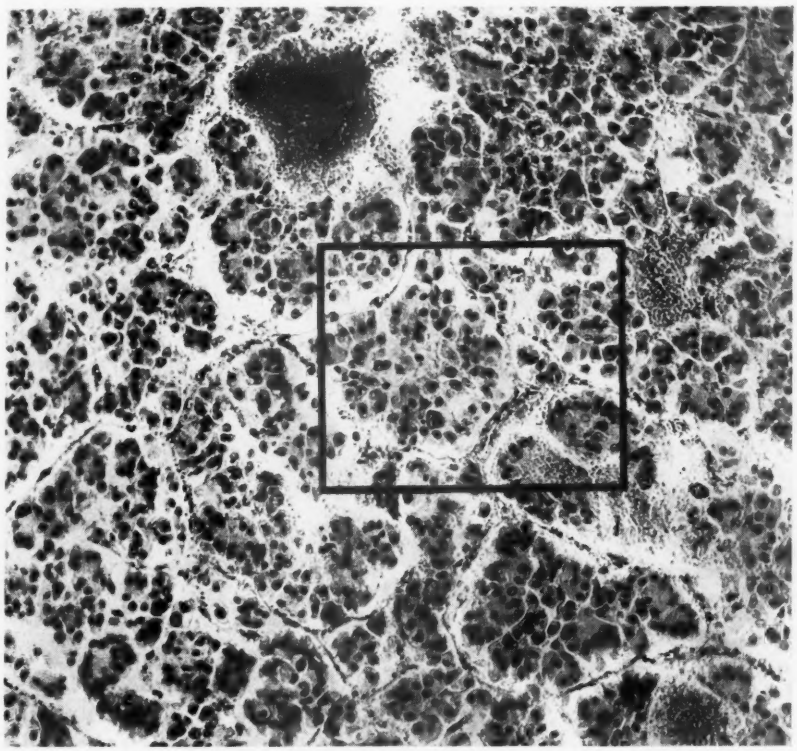


FIG. 2.—A photomicrograph of the tumor tissue in one of the metastatic nodules in the liver. The tendency toward an acinar arrangement is pronounced, although a sheet-like formation of the cells can be seen at the left. The resemblance of the tumor cells to the cells of the islands of Langerhans is striking. Areas of focal hemorrhage are present. (× 150—Hematoxylin and eosin stain)

The liver was enlarged and weighed 2500 grams. The parenchyma was brownish in color and firm in consistency. The liver was riddled throughout with whitish, circumscribed nodules of varying size, from that of a pea to that of a lime. The largest of these masses was located in the right lobe and measured seven centimeters in diameter. On the section, the larger of the nodules were found to contain a sticky, bluish-red material, which was considered as evidence of degeneration.

Both adrenals were entirely negative on section. The bowel was normal except for the area of the splenic flexure which was adherent to the tumor mass; only the serosa was involved, and the bowel wall was intact. No lymphadenopathy was found.

Microscopic.—The tumor tissue was found to be made up of sheets and nests of small polyhedral cells with pale basophilic cytoplasm. The cells varied somewhat in size and shape, and in some areas columnar cells were common. The nuclei were large and oval or round. The majority were vesicular, but many were pycnotic and hyper-

chromic. The nucleoli were prominent. Large multinucleated cells were occasionally seen. Mitotic figures were very few. In some areas the cells tended toward an acinar arrangement which was sometimes marked; in other places they were arranged in sheets or thin strands. The resemblance of the tumor cells to the cells of the islands of Langerhans was striking. The supporting stroma consisted of ill-defined and palely-staining collagenous material which was found in thin strands or, less frequently, in wide trabeculae. Many small blood vessels were seen, and focal hemorrhage and degeneration were noted.

The pancreas itself was histologically normal. The islet cells were smaller than the cells composing the tumor.

The metastatic nodules in the liver presented the same microscopic characteristics as did the parent tumor tissue (Figs. 2 and 3). The supportive stroma was more abundant in the hepatic lesions.

The microscopic examination of the other organs was essentially negative.

Attempts to demonstrate specific granules in the cells of the tumor tissue were inconclusive. Assay of the tumor tissue for insulin content was not performed.

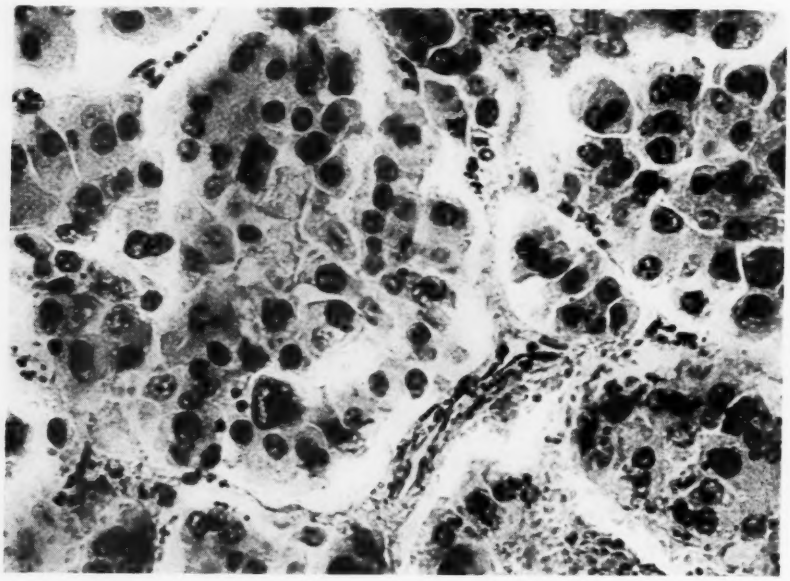


FIG. 3.—A higher-powered view of the blocked-in portion of the field shown in Figure 2. The vesicular and hyperchromic types of nuclei can be seen, and the nucleoli are prominent. (x 620—Hematoxylin and eosin stain)

DISCUSSION

An analysis of the various features presented in the series of cases of islet cell carcinoma tabulated in Table I is of both pathologic and clinical interest.

Pathologically, the size of the pancreatic masses varied considerably. The smallest tumor measured 1.8 x 1.8 x 1.3 cm.; the largest, with recorded dimensions, measured 8 x 8 x 6.5 cm., although lesions the size of a fetal head and the size of a small grapefruit have been reported.

So far as the site of the tumor in the pancreas is concerned, five of them occurred in the head, six in the tail, two in the head and body, and two in the tail and body. The entire pancreas was involved in five instances. In

no case has the body of the pancreas alone been the site of an islet cell carcinoma. Maximov and Bloom[41] state that the islands of Langerhans are somewhat more numerous in the tail of the pancreas, and Duff[16] has pointed out that islet cell adenomata are found most frequently in the tail. In the series of islet cell malignancies, however, the head and the tail have been the site of the tumor in almost an equal number of instances.

Grossly, the lesions were, as a rule, fairly well circumscribed, although in some cases surrounding adhesions and infiltration of neighboring organs have been described. The tumors were whitish or yellowish in color, and generally firm in consistency. Areas of focal hemorrhage and necrosis have been mentioned. Nodularity of the tumor mass was the rule. The metastatic lesions have presented much the same appearance grossly as did the primary pancreatic tumors.

Microscopically, the appearance has been essentially identical in all cases. The tumor tissue was composed of small polyhedral or columnar cells arranged in places in sheets, clusters, or strands, and characteristically showing a decided tendency toward acinar formation. The resemblance of the tumor cells to the cells of the island of Langerhans has been remarked upon repeatedly. Some degree of variability in the size of the cells has been present. The cytoplasm of the tumor cells has taken either a pale basic or a pale acid stain, more commonly the former. A tendency toward infiltration of the adjacent pancreatic parenchyma and cellular anaplasia of various degrees have been described. The nuclei of the cells have been vesicular or hyperchromatic. Multinucleated cells have been described in two instances (Cases 11 and 22). Mitotic figures have, as a rule, been infrequently found. In four instances staining for specific granules has been successfully accomplished (Cases 14, 15, 10 and 20). The supporting stroma has varied from delicate strands to wide bands of connective tissue. The metastatic lesions have shown, histologically, essentially the same characteristics as the parent tumor.

Metastases were widespread and generalized in five cases. The liver was involved metastatically in 18 cases, and the regional nodes in 11.

In three of the cases reported insulin assay of the tumor tissue has been successful (Cases 6, 11 and 13). With respect to the benign islet cell adenomata, successful insulin assay has been reported in at least eight instances.[47, 38, 21]

From the clinical point of view a number of interesting facts are to be gained from a survey of the cases of islet cell carcinoma reported:

Of 19 cases in which the sex of the patient could be ascertained, 14 occurred in males and five in females.

The age of the youngest case reported was 18; that of the oldest 73. The average age of the patients was 47.8 years. Six cases occurred in persons 39 or under; ten in the age-group of 40 to 60; and four in patients 61 or over.

Figures of the duration of survival after the onset of symptoms were

available in 14 of the 22 cases. The shortest period of survival was three and one-half weeks; the longest recorded was four and one-half years. Five patients lived a year or more after their symptoms began; and six died in six months or less. The average duration of life once symptoms were noted was 11.9 months.

In all of the 11 cases, where adequate clinical data was available, hypoglycemia of marked degree was present and the progressive increase in the severity and the frequency of the hypoglycemic episodes constituted the principal clinical feature. In the three cases mentioned briefly by Duff,[15] the absence of hypoglycemia was remarked upon. In the hypoglycemic cases blood sugar levels in the low twenties have not been uncommon; in one case a blood level of 16 mg. per cent was noted. According to Womack[64] "cancer of the islet cell type presents the most profound states of hypoglycemia that are encountered clinically." The clinical and pathologic aspects of hypoglycemia have been thoroughly reviewed by several authors.[60, 64, 56]

Jaundice has been noted in only two instances (Cases 1 and 2). These cases were among the group of 12 tumors which involved the head, five of which were limited to the head alone. Berk,[5] in his review of pancreatic carcinoma of the common acinar type, found jaundice in 81.3 per cent of the malignancies involving the head of the pancreas.

In two of the 11 cases, where adequate clinical data was obtained, there was a history of diabetes mellitus prior to the onset of the symptoms of hyperinsulinism (Cases 10 and 11). One of the five cases reported by Harris[26] in his original article had had glycosuria, and Herxheimer,[29] Heiberg,[27] and Smith and Seibel,[51] have reported islet cell adenomata in diabetics. It is of interest to note, in passing, that Berk,[5] in his review of the acinar variety of pancreatic malignancy, found that 6.9 per cent of the patients in this group had antecedent diabetes, whereas the incidence of diabetes mellitus in the cancer group, at large, he placed at one to two per cent.

SUMMARY

1. The literature on the subject of carcinoma of the islet cells of the pancreas has been reviewed.

2. It has been pointed out that, at present, the only valid criterion of malignancy in cases of islet cell tumor is the presence of metastases or invasion of neighboring organs, the histologic characteristics of the tumor notwithstanding.

3. Twenty-one cases of islet cell carcinoma with involvement of other structures have been culled from the literature and an additional case added. An analysis of these cases has been presented.

We are indebted to Drs. Eugene A. Case, Henry L. Bockus, and Walter Estell Lee for permission to publish the case presented and for their kind assistance in the preparation of this paper.

BIBLIOGRAPHY

1 Ballinger, J.: Hypoglycemia from Metastasizing Insular Carcinoma of Aberrant Pancreatic Tissue in the Liver. Arch. Path., **32,** 277, 1941.

2 Banting, F. G., and Best, C. H.: The Internal Secretion of the Pancreas. J. Lab. and Clin. Med., **7,** 251, 1922.

3 Banting, F. G., Campbell, W. R., and Fletcher, A. A.: Further Clinical Experiences with Insulin in the Treatment of Diabetes Mellitus. Brit. Med. Jour., **1,** 8, 1923.

4 Bast, T. H., Schmidt, E. R., and Sevringhaus, E. L.: Pancreatic Tumor with Hypoglycemic Status Epilepticus. Acta Chir. Scand., **71,** 82, 1932.

5 Berk, J. E.: Diagnosis of Carcinoma of the Pancreas. Arch. Int. Med., **68,** 525, 1941.

6 Berardinelli, W.: Hyperinsulisme et Hypoglycémie au cours d'un Adéno-Carcinome du Pancréas. Presse Med., **42,** 2098, 1934.

7 Bickel, G., Mozer, J. J., and Junet, R.: Diabéte avec dénutrition grave. Disparition de la glycosuria et atténuation progressive de l'hyperglycémie à la suite du development d'un carcinome insulaire du pancréas avec métastases hépatiques massives. Bull. et Mem. de la Soc. Méd. des Hop. de Paris., **51,** 12, 1935.

8 Bru, P.: Cancer Langerhansien généralisé chez un chien. Rev. Med.—Chir. des Maladies du Foie, du Pancréas, et de la Rate, **2,** 40, 1927.

9 Brunschwig, A.: The Surgery of Pancreatic Tumors. C. V. Mosby Co., St. Louis, 1942.

10 Campbell, W. R., Graham, R. R., and Robinson, W. L.: Islet Cell Tumors of the Pancreas. Am. J. Med. Sci., **198,** 445, 1939.

11 Cannon, P. R.: Cited by Gray.[24]

12 Cecil, R. H.: Concerning Adenomata Originating from the Islands of Langerhans. J. Exper. Med., **13,** 595, 1911.

13 Cottalorda, J., and Escarras.: Epithelioma Langerhansien: Extirpation, guérison. Lyon. Chir., **30,** 148, 1933.

14 Cragg, R. W., Power, M. H., and Lindem, M. C.: Carcinoma of the Islands of Langerhans with Hypoglycemia and Hyperinsulinism. Arch. Int. Path., **60,** 88, 1937.

15 Duff, G. L.: The Clinical and Pathological Features of Carcinoma of the Body and Tail of the Pancreas. Bull. Johns Hopkins Hosp., **65,** 69, 1939.

16 Duff, G. L.: The Pathology of Islet Cell Tumors of the Pancreas. Am. J. Med. Sci., **203,** 437, 1942.

17 Evangelisti, T.: Sui carcinomi pancreatici a cellule di tipo insulare. Policlinico (sez. chir.), **42,** 384, 1935.

18 Fabozzi, S.: Über die Histogenese des primären Krebses des Pankreas. Ziegl. Beitr. z. path. Anat. u. z. allg. Path., **34,** 199, 1903.

19 Fahri, A., and Sédad, A.: Contribution à l'étude des tumeurs malignes de la queue du pancréas. Schweiz. Med. Wochenschrift., **10,** 412, 1929.

20 Flinn, L. B., Beatty, G. A., Ginsberg, M., and Hemsath, F. A.: Carcinoma of the Islands of Langerhans with Hypoglycemia and Metastasis to the Liver. J. A. M. A., **117,** 283, 1941.

21 Frantz, V. K.: Tumors of Islet Cells with Hyperinsulinism; Benign, Malignant, and Questionable. Annals of Surgery, **112,** 161, 1940.

22 Fletcher, A. A., and Campbell, W. R.: The Blood Sugar following Insulin Administration and the Symptom Complex Hypoglycemia. J. Metab. Res., **2,** 637, 1922.

23 Gomori, G.: Observations with Differential Stains on Human Islets of Langerhans. Am. J. Path., **17,** 395, 1941, Case 4449.

24 Gray, L. M.: Functioning Islet Cell Carcinoma with Metastases to Liver. Am. J. Path., **18,** 633, 1942.

25 Hamdi, H.: Ein Insulargenetisches Pankreascarcinom (Insulom). Ztsch. f. Krebsforsch., **37,** 411, 1932.

26 Harris, S.: Hyperinsulinism and Dysinsulinism. J. A. M. A., **83,** 729, 1924.

[27] Heiberg, K. A.: Ein Fall von Adenom in den Langerhansschen Inseln der Bauchspeicheldrüse bei einem Diabetiker. Centralb. f. allg. Path. u. path. Anat., **22**, 532, 1911.

[28] Helmholz, H. F.: An Adenoma of Island of Langerhans. Bull. Johns Hopkins Hosp., **18**, 185, 1907.

[29] Herxheimer, G.: Über Pankreascirrhose (bei Diabetes). Arch. f. path. Anat. u. Physiol., **183**, 228, 1906.

[30] Howland, G., Campbell, W. R., Maltby, E. J., and Robinson, W. L.: Dysinsulinism: Convulsions and Coma due to Islet Cell Tumor of Pancreas: Operation and Cure. J. A. M. A., **93**, 674, 1929.

[31] Hueper, W. C.: Islet Adenoma in the Pancreas of a Mouse. Arch. Path., **22**, 220, 1936.

[32] Joachim, H., and Banowitch, M. M.: A Case of Carcinoma of the Islands of Langerhans with Hypoglycemia. Ann. Int. Med., **11**, 1754, 1938.

[33] Jacobsen, V. C.: Carcinoma of the Islets of Langerhans. Arch. Path., **18**, 135, 1934.

[34] Joslin, E. P., Root, H. F., White, P., and Marble, A.: The Treatment of Diabetes Mellitus. Lea and Febiger, Philadelphia, 1940, p. 335, Case 2.

[35] Judd, E. S., Allan, F. N., and Rynearson, E. H.: Hyperinsulinism: Its Surgical Treatment, J. A. M. A., **101**, 99, 1933.

[36] Judd, E. S., Faust, L. S., and Dixon, R. K.: Carcinoma of the Islands of Langerhans with Metastasis to the Liver Producing Hyperinsulinism. West. J. Surg., Obs., and Gyn., **42**, 555, 1934.

[37] Keating, F. R., and Wilder, R. M.: Spontaneous Hypoglycemia: Report of Cases. South. J. Med. and Surg., **103**, 125, 1941.

[38] Kerwin, A. J.: Fatal Hyperinsulinism with Cerebral Lesions due to Pancreatic Adenoma. Am. J. Med. Sci., **203**, 363, 1942.

[39] Lang, F. J.: Über einige Geschwulstbildungen des Pankreas. Arch. f. path. Anat. u. Physiol., **257**, 235, 1925.

[40] Lloyd, P. C.: A Case of Hypophyseal Tumor with Associated Tumor-like Enlargement of the Parathyroids and Islands of Langerhans. Bull. Johns Hopkins Hosp., **45**, 1, 1929.

[41] Maximow, A. A., and Bloom, W.: A Textbook of Histology. W. B. Saunders Co., Philadelphia, 2d ed., p. 432, 1936.

[42] Meyer, K. A., Amtman, L., and Perlman, L.: Islet Cell Tumors of the Pancreas: Report of a Case. J. A. M. A., **117**, 16, 1941.

[43] Moersch, F. P., and Kernohan, J. W.: Hypoglycemia: Neurologic and Neuropathologic Studies. Arch. Neurol. and Psych., **39**, 242, 1938.

[44] Morse, M. E.: Two Adenomata of the Islands of Langerhans. J. A. M. A., **51**, 1075, 1908.

[45] Nicholls, A. G.: Simple Adenoma of the Pancreas Arising from an Island of Langerhans. J. Med. Research, **8**, 385, 1902.

[46] Pappenheimer: Cited by Whipple and Frantz.[59]

[47] Quarrier, S. S., and Bingham, C. T.: Adenoma of the Pancreas. ANNALS OF SURGERY, **115**, 363, 1942.

[48] Rudd, T. N., and Walton, J.: A Case of Islet Cell Adenoma of the Pancreas. Brit. J. Surg., **29**, 266, 1942.

[49] Seckel, H. P. G.: Postmortem Hepatic Glycogenolysis in Hyperinsulinism and Glycogen Diseases. J. Clin. Invest., **18**, 723, 1939, Case 195034.

[50] Slye, M., and Wells, H. G.: Tumors of Islet Tissue with Hyperinsulinism in a Dog. Arch. Path., **19**, 537, 1935.

[51] Smith, M. G., and Seibel, M. G.: Tumors of the Islands of Langerhans and Hypoglycemia. Am. J. Path., **7**, 723, 1931.

52 Thalhimer, W., and Murphy, F. D.: Carcinoma of the Islands of Langerhans: Hyperinsulinism and Hypoglycemia. J. A. M. A., **91,** 89, 1928.

53 Vecchio, A.: Adenoma maligno delle isole di Langerhans di un pancreas aberrante. Arch. per le sc. med., **38,** 277, 1914.

54 Warren, S.: Adenomas of the Islands of Langerhans. Am. J. Path., **2,** 335, 1926.

55 Warren, S.: The Pathology of Diabetes Mellitus. Lea and Febiger, Philadelphia, 1930, p. 181.

56 Wauchope, G. M.: Hypoglycemia. Quart. J. Med., **2,** 117, 1933.

57 Whipple, A. O.: The Surgical Therapy of Hyperinsulinism. J. Internat. de Chir., **3,** 237, 1938.

58 Whipple, A. O.: Present-Day Surgery of the Pancreas. New Eng. J. Med., **226,** 515, 1942.

59 Whipple, A. O., and Frantz, V. K.: Adenoma of Islet Cells with Hyperinsulinism. ANNALS OF SURGERY, **101,** 1299, 1935.

60 Wilder, R. M.: Spontaneous Hypoglycemia. Internat. Clin., **3,** 143, 1936.

61 Wilder, R. M., Allan, F. N., Power, M. H., and Robertson, H. E.: Carcinoma of the Islands of the Pancreas: Hyperinsulinism and Hypoglycemia. J. A. M. A., **89,** 348, 1927.

62 Willis, R. A.: A Rare Type of Diffuse Carcinoma of the Pancreas with Unusual Metastases. J. Path. and Bact., **42,** 203, 1936.

63 Winters, W. L., Gottardo, P., and McNealy, R. W.: Severe Hypoglycemia due to Islet Adenoma of the Pancreas: Surgical Cure. W. J. of S. G. and O., **49,** 488, 1941.

64 Womack, N. A.: Hypoglycemia. Surgery, **2,** 793, 1937.

65 Zanetti, G.: Contributo allo studio dei tumori del pancreas. Arch. per le sc. med., **49,** 505, 1927.

CHOLESTEROSIS OF THE GALLBLADDER*

OBSERVATIONS ON TWENTY-FIVE CASES WITHOUT STONES

KENNETH M. LEWIS, M.D., AND CHARLES W. PETERSON, M.D.

NEW YORK, N. Y.

FROM THE FOURTH SURGICAL DIVISION, BELLEVUE HOSPITAL, NEW YORK, N. Y.

DURING THE PAST 12 YEARS, 455 patients suffering from gallbladder disease have been operated upon on the Fourth Surgical Division at Bellevue Hospital. The cases have been studied preoperatively and followed postoperatively in a special Gallbladder Clinic. It is felt that a clinic of this type affords greater opportunity for more careful study and follow-up of these patients. In this group there were a certain number of patients whose clinical and laboratory data were not entirely conclusive, and where operation was undertaken with some indecision as to the operative findings. These latter patients frequently were found to be suffering from cholesterosis of the gallbladder, but preoperative recognition of the condition was often most difficult.

Cholesterosis of the gallbladder may be defined as a metabolic and not an inflammatory disease in which the mucosa of the gallbladder contains deposits of cholesterol and other lipoid material. In the advanced stages of the disease cholesterol calculi may be present in the lumen of the gallbladder. Virchow,[1] in 1887, first noted lipoid deposits in the gallbladder, and Moynihan,[2] in 1919, in noting yellow stippling of the gallbladder mucosa, first brought cholesterosis of the gallbladder into surgical prominence by describing it as a "condition of the gallbladder requiring cholecystectomy." MacCarty,[3] in 1910, in describing the advanced stage of the disease originated the term "strawberry gallbladder," and since that time numerous authors have reported both on the clinical and research aspects of the condition. Whether the cholesterol enters the gallbladder wall from the bile (absorption), as suggested by the experiments of Illingsworth,[4] and of Rousselot and Bauman,[5] or from the blood (excretion), as claimed by Naunyn,[6] and Elman and Graham,[7] seems, clinically, to be of little consequence. The entire subject has been brought carefully up to date in the splendid analysis by Mackey,[8] in 1937.

It is the purpose of this communication to report 25 cases of cholesterosis without stones, in which the diagnosis of a diseased gallbladder at the operating table was frequently most difficult, and to suggest an added aid in diagnosis, namely, biliary drainage, which, so far, has not been reported in the literature as a diagnostic method in the recognition of this disease. Of the 455 patients with proven gallbladder disease operated upon, 40 had cholesterosis of the gallbladder confirmed by pathologic examination. Of

* Read before The New York Surgical Society, November 25, 1942.

these 40 cases, 15 had calculi and these cases are excluded, as diagnosis at the operating table of gallbladder disease was self-evident by the palpation of calculi within the gallbladder.

In the remaining 25 cases calculi were not present, the pathology ranged from a few cholesterol plaques to a well-defined strawberry gallbladder, and diagnosis of a pathologic viscus at the time of operation by palpation and inspection of the gallbladder was extremely difficult. It is in this group that any preoperative aid in diagnosis seemed most desirable.

Of the 25 patients 20 were women and five were men, showing the large predominance of the female over the male in this condition. Eighteen of the patients were markedly obese, which would fit in well with the metabolic character of the disease. Twenty-one of the patients had complaints of indigestion, notably, flatulence, belching, epigastric distress after meals, nausea and vomiting, but the degree and number of the digestive complaints varied considerably. Three of the patients who had no symptoms of indigestion complained only of attacks of right upper quadrant colicky pain. The fourth patient who had no digestive complaints noted persistent dull aching pains in the gallbladder region but had no attacks of colic. Operation in this particular patient was postponed for almost two years, as the diagnosis of gallbladder disease did not seem certain enough to warrant surgery.

Of the 21 patients who complained of one or more of the symptoms of indigestion, 15 noted that fatty foods aggravated their symptoms, while in the remaining six, the digestive disturbances were increased by the ingestion of all types of food.

TABLE I
PREOPERATIVE SYMPTOMS—SEX—AND PHYSICAL STATUS

No. of Cases	Sex	Obese	Not Obese	Indigestion	Pain	
					Colic	Dull Aching
25	20 Females 5 Males	18	7	21	19	6

Pain was a constant factor in all of the cases, and, in analysing the group, it was surprising to note how many of the patients complained of recurrent attacks of colicky pain in the right upper quadrant of the abdomen radiating to the right scapula or right shoulder. Eighteen patients complained of frequent attacks of this type of colicky pain, one patient noted it only infrequently, and six patients complained only of a dull aching pain, with no attacks of colic. Dr. John H. Morris[9] in a paper read before The New York Surgical Society, in May, 1934, reported on 14 cholesterosis patients, and found that attacks of biliary colic were present in 13 of the 14 cases.

Tenderness in the gallbladder region was noted in all of the patients at one or more examinations. This seemed to be a constant finding, although it was not always present at every examination. It was always noted when the patient was seen in an attack of colicky pain.

Blood cholesterol estimations were obtained preoperatively, and two to three months postoperatively, on 16 of the 25 patients. The preoperative blood cholesterol figures ranged from 125 to 235 mg. per cent, and showed such a wide divergence that no conclusions could be drawn therefrom

(Table II). Postoperatively, the blood cholesterol ranged from 125 to 180 mg. per cent, showing a definite drop in only three of the patients. It would thus seem as though blood cholesterol estimations were of little diagnostic aid, and this would conform to the findings of previous observers.

TABLE II

BLOOD CHOLESTEROL ESTIMATIONS

	No. of Cases	125-150 mg. per cent	150-175 mg. per cent	175-200 mg. per cent	200-235 mg. per cent
Preoperative	16	7	4	2	3
Postoperative	16	8	3	5	0

Cholecystographic roentgenologic studies in this series were very inconclusive. Fifteen of the patients showed diminished visualization with delayed emptying, five showed normal visualization with good emptying, and only five showed no visualization with the dye. Thus, in 20 of the patients cholecystograms were not conclusive as a diagnostic procedure. It is always difficult to evaluate a diminished or faint visualization, or to properly correlate delayed emptying by the gallbladder shadow.

It has been our custom to, preoperatively, do a biliary diagnostic drainage on all patients with suspected disease of the gallbladder. If a satisfactory drainage is not obtained at the first attempt, the drainage is repeated on one or more occasions until adequate samples of bile are secured. These samples are then allowed to stand in the icebox for from 24 to 48 hours, permitting any solid material in the bile to settle out. Microscopic examination of any sediment drawn up with a pipette is then made.

In studying the above patients with cholesterosis of the gallbadder one very frequent finding has been noted. In 19 of the 25 cases cholesterol crystals in the "B" drainage bile has been found in readily recognizable amounts. Concentrated dark green "B" bile was obtained in all of the patients, but calcium bilirubinate pigment was found in only three instances. It is extremely important that concentrated "B" bile be obtained. This is the bile contained in the gallbladder, and it is in this specimen that a search for cholesterol crystals should be made. Examination of samples of yellow bile obtained from the common bile duct may not show the crystals. If a specimen of "B" bile is not obtained at any one drainage, the drainage should be repeated at a subsequent time. It would thus seem to be suggestive that with cholesterol crystals seen in the drainage bile, and with inconclusive cholecystographic findings, a diagnosis of cholesterosis of the gallbladder might justifiably be entertained.

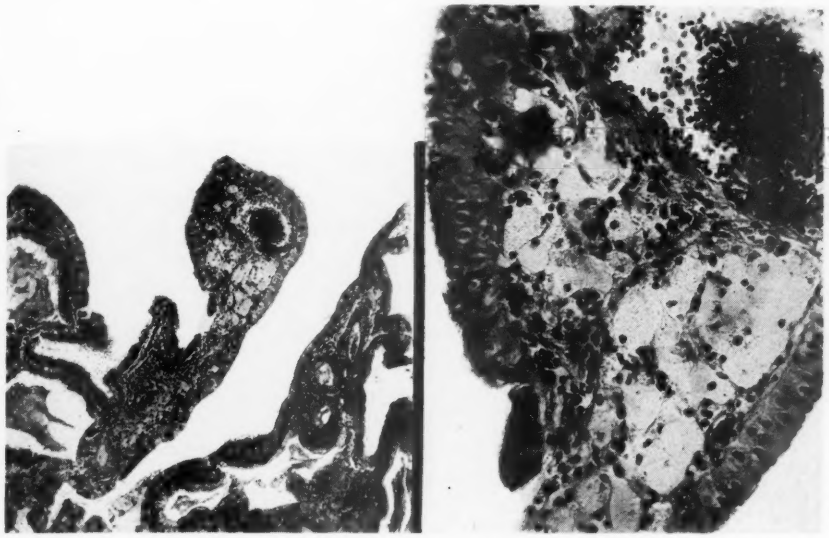
TABLE III

BILIARY DRAINAGE DATA

No. of Cases	Cholesterol Crystals	Calcium Bilirubinate	"B" Bile
25	19	3	25

One of the greatest difficulties in this disease has been the recognition of any real pathology of the gallbladder at the operating table. This of course applies to those cases without calculi. The gallbladder wall is thin, translucent, and not thickened. The serosa of the viscus has its normal sheen, and usually no adhesions to neighboring viscera are found. Palpation of the

gallbladder reveals little. It empties readily, is usually not enlarged or contracted, and thickening of the wall cannot be felt. The bluish sheen that has been described by some observers has not proved reliable in the hands of the authors. In other words, the patient has been operated upon with a preoperative diagnosis of gallbladder disease, and at the operating room table an apparently normal gallbladder is found. Nothing can be



A B

FIG. 1.—A. and B.: Cholesterosis of the Gallbladder. Note the distended, foamy cells, containing lipoid in the villi of the mucous membrane. These are seen as the pale areas in the stroma. (Paraffin section: Hematoxylin-eosin—(A) Low power and (B) high power).

more disturbing than to decide whether to remove an apparently normal viscus or to leave it in. Two identical instances as described above were encountered recently. Because cholesterol crystals had been found in the preoperative drainage bile, it was decided to perform a cholecystectomy in both patients, even though the gallbladder, from the serosal surface, seemed normal. Both gallbladders, after removal, showed numerous early cholesterol and lipoid deposits in the mucosa. These could be recognized grossly and were subsequently confirmed by pathologic examination.

In another instance, where, unfortunately, a preoperative biliary drainage had not been done, and where the gallbladder appeared grossly normal, it was assumed that the symptoms must be due to some other organ, and the gallbladder was not removed. Five days postoperatively the patient had a typical attack of biliary colic. Two months later a cholecystectomy was performed and a typical early cholesterosis of the gallbladder was found.

Our routine surgical procedure has been cholecystectomy, and this was performed on all of the 25 patients here reported. There was no mortality in this group, all of the patients making a satisfactory operative recovery.

We were able to follow 20 of the patients in this series. Two of these have been followed from one to two years, three from two to three years,

three from three to four years, and the remaining twelve from five to ten years. The end-results of cholecystectomy have been extremely gratifying. Of the 20 patients followed, 14 considered themselves completely cured of their previous complaints. They were able to eat all types of food with impunity and their attacks of pain disappeared. Four patients remained symptom-free as long as they stayed on a low fat diet, but otherwise had recurrence of their symptoms of indigestion. Two patients noted no improvement in their symptoms, and were unrelieved by the cholecystectomy.

It might be noted that we endeavor to keep these patients on a rigid fat-free diet for a period of six months to one year following operation. The diet is then gradually returned to a normal fat intake over a period of two to three months. The patients, if very obese, are also followed in the Obesity Clinic, where attempt is made to bring their weight somewhere near normal.

Whether rigid dietary control for some time following the cholecystectomy has played a part in relieving these patients of their symptoms, cannot, of course, be stated with certainty. In some of the patients who were observed for a number of months preoperatively, however, dietary restriction of fatty foods was insisted upon, but symptoms were never completely relieved by this alone.

TABLE IV

END-RESULTS AFTER CHOLECYSTECTOMY IN 20 CASES OF CHOLESTEROSIS, WITHOUT STONES

No. of Cases	Cured	Improved (Symptom-Free on a Low Fat Diet)	Unimproved
20	14	4	2

In reviewing the literature, one noted immediately the varying results obtained by different observers in cholesterosis gallbladder patients treated by cholecystectomy. Moynihan[2] considered his six patients completely cured. Illingsworth[4] states that "treatment by cholecystectomy is the most rational procedure and appears to yield satisfactory results." Judd and Mentzer,[10] commenting on 1,000 patients and cholesterosis treated by cholecystectomy at the Mayo Clinic, felt that good results only were obtained if pain had been an outstanding symptom. Young,[11] in 1928, reported 45 patients with strawberry gallbladders, and felt that cholecystectomy gave the most marked relief. Kopp[12] in 1929 noted that seven out of eleven cases of cholesterosis without stones were completely relieved of their symptoms by cholecystectomy. Morris[9] reported seven cures out of the nine patients he was able to follow. Mackey,[8] however, in a series of 33 cholesterosis cases, without stones, found only 14 cured and two improved by cholecystectomy. White and Riddick[13] felt that cholesterosis was only a part of a general metabolic dysfunction and that cholecystectomy could not, therefore, promise relief. Stanton,[14] in 1932, had four out of six cases of cholesterosis, without stones, that were not benefitted by cholecystectomy.

It can thus be seen that there is no uniformity of opinion as to the benefits of cholecystectomy in this disease, and it is extremely difficult to rationalize the divergent results obtained by so many careful and dependable observers.

It is our feeling, however, based upon our results with the group of

cases reported herein, that cholecystectomy offers relief to most of the patients with cholesterosis of the gallbladder, and this has been noted in a number of cases who have been studied and followed for a long period of time.

In contemplating surgery in this condition it should be remembered that the disease itself never causes death. Cholecystectomy, on the other hand, is accompanied by a certain risk if the number of patients operated upon is large enough. Each patient should, therefore, be carefully observed and cholecystectomy should be reserved for those cases in whom the symptoms are so severe as to warrant surgical intervention. It is probable that a certain number of patients with this disease never progress to calculus formation. In our series, 15 out of 40 cases did have calculi. It is our impression that the number of cholelithiasis patients with accompanying cholesterosis of the gallbladder wall is probably greater than has previously been thought. Pathologic sections should be taken from different areas of all removed gallbladders, as there may be areas of cholesterosis and chronic cholecystitis in the same gallbladder. This has been noted several times in our series.

SUMMARY

1. Twenty-five cases of cholesterosis of the gallbladder without stones, treated by cholecystectomy, are reported. Twenty of these patients were followed, postoperatively, for from one to ten years.

2. Pain was a predominant symptom in this group, and 19 of the patients complained of recurrent attacks of typical biliary colic in the upper abdomen.

3. Cholecystograms were inconclusive, only five of the cases showing nonvisualization of the gallbladder.

4. Preoperative diagnostic biliary drainage revealed cholesterol crystals in the bile in 19 of the cases. This procedure is urged as a diagnostic aid in this disease, where frequently the signs, symptoms and roentgenologic findings may be so indefinite.

5. In our series cholecystectomy gave satisfactory results. Of the 20 patients whom we were able to follow, 14 were relieved of all symptoms, four were relieved as long as they stayed on a low fat diet, while two patients were unimproved.

REFERENCES

1 Virchow, R.: Virchow's Arch., **2**, 574, 1857.
2 Moynihan, B. G. A.: Annals of Surgery, **50**, 1265, 1909.
3 MacCarty, W. C.: Annals of Surgery, **51**, 651, 1910.
4 Illingsworth, C. F. W.: Brit. Jour. Surg., **17**, 203, 1929.
5 Rousselot, L. M., and Bauman, L.: Surg. Gynec. & Obst., **61**, 585, 1934.
6 Naunyn, B.: Die Entstehung u. d. Aufbau. d. Gallensteine, 1923, Jena.
7 Elman, R., and Graham, E. A.: Arch. of Surg., **24**, 14, 1932.
8 Mackey, W. A.: Brit. Jour. Surg., **24**, 570, 1937.
9 Morris, J. H.: Paper read before the New York Surgical Society, May, 1934.
10 Judd, E. S., and Mentzer, S. H.: Collected Papers of the Mayo Clinic, **19**, 310, 1927.
11 Young, E. L.: New Eng. Jour. Med., **198**, 720, 1928.
12 Kopp, J. G.: Arch. f. Klin. Chir., **151**, 411, 1928.
13 White, C. S., and Riddick, A. L.: Southern Med. Jour., **21**, 857, 1928.
14 Stanton, E.: Am. Jour. Surg., **18**, N.S., 246, 1932.
15 Boyd, W.: Brit. Jour. Surg., **10**, 337, 1922–3.

LATE INVASION OF BLADDER AND PROSTATE IN CANCER OF THE RECTUM OR RECTOSIGMOID FOLLOWING ABDOMINO-PERINEAL RESECTION*

GORDON D. OPPENHEIMER, M.D
NEW YORK, N. Y.

FROM THE UROLOGIC AND SURGICAL SERVICES OF THE MT. SINAI HOSPITAL, NEW YORK, N. Y.

THIS STUDY was undertaken after a successful palliative urologic procedure had been performed upon a patient with recurrence of the tumor in the prostrate and bladder neck seven years after an abdomino-perineal resection for cancer of the rectum. Eight additional instances of late postoperative lower urinary tract recurrence were found in the recent hospital records and form the basis of this report. Two of these nine cases were operated upon at other institutions. Three were ward cases and four were private patients operated upon by three different surgeons.

Until recently this type of case was not seen by our Urologic Service. Apparently, when vesical neck obstruction and hematuria became prominent symptoms, the patient was considered hopeless and referred for chronic institutional care. However, with improvement in the mortality rates of radical extirpation of the rectum, and with the increase in the total number of operations performed, more patients are living following the procedure, resulting in a greater interest and attention directed to the postoperative complications and sequelae.[1] Postoperative difficulties in micturition due to disturbed neurogenic vesical control and other factors have been noted and studied by many workers. We, as have others, have obtained satisfactory results in relieving bladder stasis by transurethral resection of minimal vesical neck obstruction in this type of vesical detrusor weakness. There have been several such cases in the last three years. True prostatic fibro-adenomatous obstruction has not infrequently become symptomatic after abdomino-perineal resection and has been cured similarly by transurethral resection. It is because of these good results that cases with urinary symptoms have been referred to the urologic surgeon, thus affording us the opportunity of seeing patients who had late neoplastic infiltration of the bladder and prostate.

Because of the close anatomic relationship between rectum or lower sigmoid colon and the lower urinary tract, direct involvement of the latter by the lower bowel cancer theoretically should occur with some frequency. In an "acute hospital" such as Mt. Sinai we do not see many cases presenting the terminal stages of cancer of the rectum. Through the courtesy of Dr. D. Marine, Chief of the Laboratory Division of the Montefiore Hospital, New York City, I reviewed 50 consecutive recent

* Read before the New York Surgical Society, January 27, 1943.

autopsy protocols from that hospital in which cancer of the rectum was one of the diagnoses. Ten of these patients (six females and four males) had an abdomino-perineal or a posterior resection. Of the males, one patient had involvement of bladder, prostate and seminal vesicles, and in another, prostate and seminal vesicles were infiltrated. Four of the six females had bladder involvement. The forty other patients had either no operation or palliative colostomy. Some had received radiotherapy without apparent effect. Thirteen were females and four of these had bladder involvement. Twenty-seven in this "nonextirpative" group were males. Fourteen of these, or a little over 50 per cent, had invasion by the growth of bladder, or prostate or seminal vesicles either separately or together. Twelve of the 14 had bladder involvement. Briefly, of 50 autopsied patients who were cared for in the last stages of their disease (20 per cent had a rectal resection) there was invasion of the bladder in 21 cases. Three patients in the unoperated group had rectovesical fistulae, and one a rectovaginal fistula. The end-picture of the cases with bladder involvement was essentially the same whether the rectum had been extirpated or not, namely the picture of a "frozen" pelvis. Bladder, prostate and seminal vesicle involvement in the male, and bladder and uterine involvement in the female, apparently takes place by direct extension.

Although well known, and previously reported,[2] the fact that these patients very frequently died from upper urinary tract complications, was forcibly brought to my attention by the study of these necropsies. Six of the ten cases with rectal extirpation, and 16 of the remaining 40 inoperable cases, making a total of 22 cases out of the 50, or 44 per cent, showed evidences of compression of one or both ureters in the pelvis with or without direct invasion. Most times direct invasion was not present. Several other patients had true ureteral metastases. As a result of the ureteral compression, dilatation of the ureter, renal pelvis and calices was present, often with infection, and evidence of pyelonephritis, and sometimes with multiple renal abscesses, and even perinephric suppuration. Upper urinary tract infection was either a direct or contributing cause of death in a considerable proportion of these cases. In fact, it seemed that the mortality from ureteral blockage, with renal damage and infection, would have been even greater except that some of these patients died as result of complications associated with their metastases or, rarely, from localized infection associated directly with the primary lesion. Incidentally, 33 of the 50 cases (61 per cent) had metastases; these were usually multiple.

In general, my findings are similar to those of Kickham and Bruce,[2] who, reporting from a hospital devoted entirely to the care of cancer patients, analyzed a postmortem group of 88 cases of cancer of the rectum, clinically inoperable, in whom no extirpative surgery was performed. Seventy per cent of these cases were males, and in 31.5 per cent of the males, some cancerous invasion of the prostate could be demonstrated. Forty per cent

of the entire group showed vesical involvement. Rectovesical fistulae were noted in two cases. Forty-seven point seven per cent of the 88 cases showed occlusion of one or both ureters by actual compression or direct invasion, with resulting hydronephrosis, pyelonephritis and advanced renal damage. Incidentally, renal metastases were found in two of their patients.

Metastasis to the lower urinary tract as one of multiple metastases theoretically may occur. Solitary metastasis to the bladder must be very rare, but if present in an accessible location may be successfully treated by surgical means. As judged by the postmortem findings mentioned above, all the evidence indicates that residual cancer with subsequent contiguous direct extension accounts for the late malignant involvement of the bladder, prostate, or seminal vesicles under consideration. One might state that the more advanced the primary rectal tumor is in its local growth, the greater is the likelihood that the patient will have secondary invasion of the bladder or prostate following the extirpation.

The following report is probably typical of many of the cases who are operated upon for a locally advanced lesion.

ILLUSTRATIVE CASE REPORTS

Case 1.—Hospital No. 447541: J. Z., a 49-year-old male, entered the Mt. Sinai Hospital, February 27, 1939, and was discharged, April 12, 1939. He had suffered from bilateral apical pulmonary tuberculosis 25 years before. He complained of blood in the stool for one year, with increasing frequency of defecation, rectal tenesmus and loss of weight. The report of the biopsy specimen taken from a fungating cauliflower rectal mass was infiltrating adenocarcinoma. On March 6, 1939, a one-stage abdominoperineal resection was performed. No intra-abdominal metastases were found but there was considerable fixation to the prostate. The specimen showed a circumscribed tumor, about 3.5 cm. in diameter and about 3 cm. above the anal margin, which grossly infiltrated all coats of the rectum and invaded the perirectal fat. The pathologic report was "ulcerative infiltrating adenocarcinoma of rectum. Invasion of perirectal tissue. No involved lymph nodes were found." The patient was well for three months, gained weight, but then began to complain of dysuria, frequency, and back pain. Roentgenograms of the dorsolumbar spine were negative for metastases. Six months after operation he complained of the same symptoms, with an episode of hematuria. A short course of radiotherapy was given, but perineal, back and bladder pain with frequency and nocturia continued. Because of a stricture at the site of colostomy, the colostomy was revised. He finally was admitted to the Montefiore Hospital, December 28, 1939, and died, March 29, 1940, about one year after the original operation. The postmortem findings were as follows:

Autopsy.—Local recurrence of carcinoma of the rectum, with invasion of pelvic tissues, pubic bones, bladder, prostate and seminal vesicles and with metastases to the liver, ureters, vertebrae, regional and celiac lymph nodes. Bilateral ureteropyonephrosis, with suppurative pyelonephritis. Chronic fibroid pulmonary tuberculosis, with cavitation. Bilateral bronchopneumonia. Bronchiectasis upper lobes. Chronic cystitis, with incrustations.

Comment: The patient was operated upon one year after symptoms had commenced. Undoubtedly, cancer cells were left on the posterior aspect of the prostate and bladder and in the perirectal lymphatics and fat. Local

recurrence and involvement of the lower urinary tract became apparent. No surgical or urologic therapy could have been of any value. He died with metastases, urosepsis, and terminal bronchopneumonia.

Contrasted with this is the unusual case mentioned in the introduction of this paper.

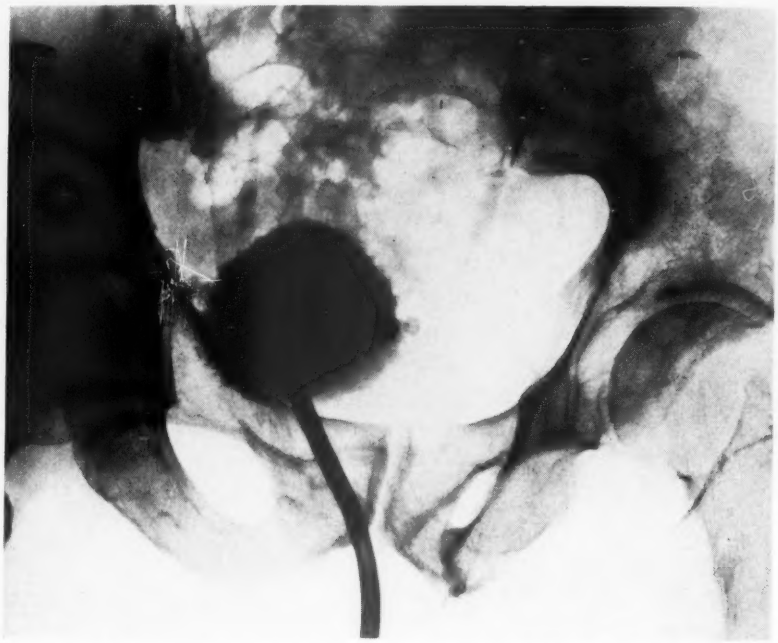


FIG. 1.—Cystogram. Small diverticulated bladder on right side of pelvis. Bladder elevated above pubis. Normal lower end of ureters visualized by excretory urogram.

Case 2.—Hospital No. 477773: H. A., a 66-year-old man, was admitted to the Mt. Sinai Hospital, August 16, 1941, and discharged, August 26, 1941. Seven years previously, August 17, 1934, a one-stage combined abdomino-perineal resection had been performed. (Dr. D. Jones, Palmer Memorial Hospital, Boston, Mass.) The growth was annular, started about 12 cm. above the anus and was 2.5 cm. wide. It was situated above the peritoneum, which covered a very deep rectovesical pouch. The neoplasm penetrated the entire wall of the bowel and had grown through the peritoneum on the anterior surface. There were nodules on the adjacent peritoneum, and the tumor had apparently extended through to involve the seminal vesicles. There were no liver metastases. At the second part of the operation, when the ischiorectal fat, sphincter, rectum and growth were removed from below, the surgeon commented on the technical difficulty because of the seminal vesicle involvement. The seminal vesicles were removed with the tumor and the bowel. The pathologic report was "malignant adenoma, with metastases to one lymph node." Grossly, the seminal vesicles were infiltrated by tumor tissue. The prognosis was considered poor. The patient was given radiotherapy by Dr. J. R. Freid, of this city. Two years after operation, a hard, fixed mass developed in the right lower quadrant of the abdomen but disappeared after intensive radiotherapy.* He was well until eight months before admission to this

* Details of Radiotherapy (Dr. J. R. Freid): From 1/18/35 to 2/1/35, and from 5/16/35 to 6/7/35, eleven small X-ray treatments, totaling 1800 r. high voltage to perineal sinus. 7/19/37 to 8/20/37, treated over 15x14 cm., anterior and posterior right lower pelvic portals, each of these areas receiving 3000 r. Factors were 200 K.V., Thoreus filter (equal to 2 mm. Cu.) 80 cm. T.S.D.

institution, when he suffered from gradually increasing frequency of urination and burning on urination. Examination revealed a fair state of nutrition, but some evidence of weight loss, a well functioning colostomy to the left of the umbilicus, a satisfactory perineal scar, and systolic cardiac murmurs at the aortic and mitral areas. The residual urine was six ounces. The urine contained pus and *B. coli*. Blood urea nitrogen was 10 mg. per 100 cc. An excretory urogram showed normal excretion on both sides, without dilatation of the pelves, calices or ureters. The latter were visualized to the bladder. The cystogram (Fig. 1) indicated a diverticulated bladder, which

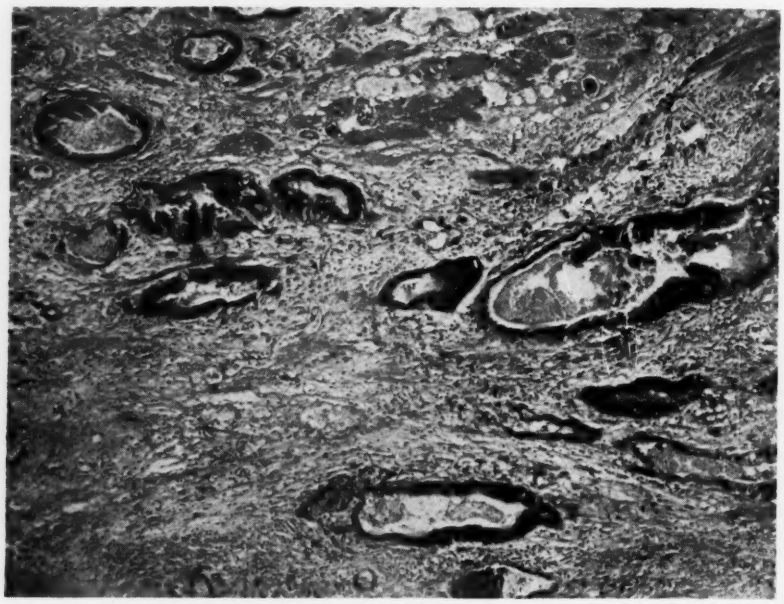


FIG. 2.—Photomicrograph of transurethrally resected tissue showing infiltrating adenocarcinoma similar to original rectal carcinoma.

appeared to be fixed to the right side of the pelvis. A preliminary bilateral vas transection was performed. On August 8, 1941, under low spinal anesthesia, a cystoscopy and transurethral resection were performed. The bladder was trabeculated. There was intra-urethral lateral lobe encroachment both in the midline and superiorly. The inferior vesical sphincter was elevated. On the left side some of the resected tissue was spongy and different from the right side. Fifteen grams of tissue were removed. The patient was discharged five days later, voiding well, without residual. The pathologic report (Dr. P. Klemperer) was adenocarcinoma infiltrating the prostate (Fig. 2). A comparison with a slide of the original specimen led Dr. Klemperer to state that they were quite identical histologically, and that the cancer in the transurethrally resected tissue was definitely secondary to direct extension from residual rectal growth. He received further very intensive high voltage radiotherapy,* and was well up to November, 1942 (15 months), except for a short episode of urinary frequency and burning, with pyuria, which promptly responded to sulfadiazine therapy. There was no residual urine.

* From 9/2/41 to 11/4/41, treated over five fields: right and left pubic, right and left gluteal, and perineal portal. He received 2500 r. to each of these areas. Other factors were 200 K.V., filtration 0.5 mm. Cu. plus 3 mm. Al., 80 cm. T.S.D.

On November 16, 1942 (eight years and three months after abdomino-perineal resection) the patient was readmitted to the Mt. Sinai Hospital because of recurrent urinary frequency, dysuria, urgency, and pyuria. A large vesical calculus was seen roentgenologically (Fig. 3). The excretory urogram, again, indicated a normal upper urinary tract, and, again, the bladder (Fig. 4) was situated on the right side of the pelvis but showed no prostatic intrusion. Under spinal anesthesia, the stone was crushed and evacuated. The vesical outlet and prostatic urethra presented only a very small amount of intruding tissue—from this area three grams of tissue were removed with the McCarthy resectoscope, for biopsy. Much to my surprise, none of this tissue showed any evidence of cancerous infiltration. The serum "acid" phosphatase was four King-Armstrong units. The patient was discharged several days later, and has been well since.

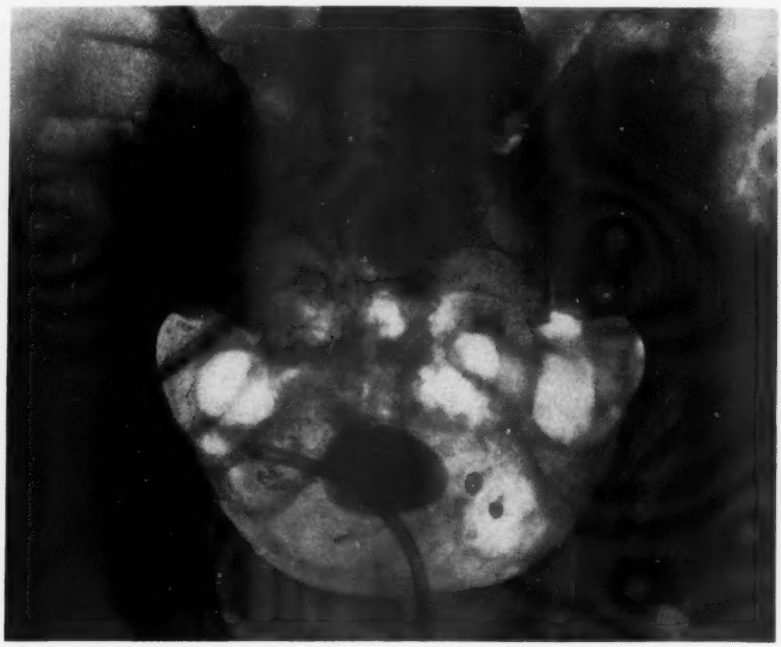


FIG. 3.—Large vesical calculus. Two smaller shadows are outside of bladder.

Comment: Histologically, the original rectal tumor and the infiltrating neoplasm affecting the prostrate were similar. Biologically, however, this tumor is unusual because it is very slow growing and evidently radiosensitive. The result obtained justifies our frequent attempts to undertake radical surgery even though the local lesion is fixed to surrounding structures and also justifies later palliative therapeutic efforts.

Case 3.—Hospital No. 484107: B. W., a 49-year-old physician, had three admissions to the Mt. Sinai Hospital, the first in May, 1941. His past urologic history was that of right renal colic in 1911, and, in 1917, colic with the spontaneous passage of a stone. Four years before admission, a one-stage abdomino-perineal resection with a left iliac colostomy had been performed at another institution for a cancer of the rectosigmoid. He had been well until shortly before admission, when frequency, dysuria, loin pain, and painless hematuria developed. Cystoscopy showed a solid spher-

ical-shaped neoplasm about the size of a hickory nut on the trigone, to the left of the right ureter orifice and extending towards the left lateral wall. The pathologic report of the biopsy was "fragments of adenocarcinoma." On May 20, 1941, an exploratory celiotomy was performed (Dr. A. Hyman). The peritoneum was free of metastases. The liver and pelvic region showed no gross evidence of neoplasm. The bladder was mobilized and opened in the midline. A flat necrotic growth, the size of a quarter, was found in the bladder, overlying the right ureter orifice and extending close to the sphincter. The base of the bladder was indurated. The tumor was treated by the introduction of 12 platinum radon seeds of 0.75 millicuries each, placed circularly around the periphery and center of the growth. Suprapubic Malecot catheter drainage was established. One month later, the tumor was fulgurated transurethrally. The supra-

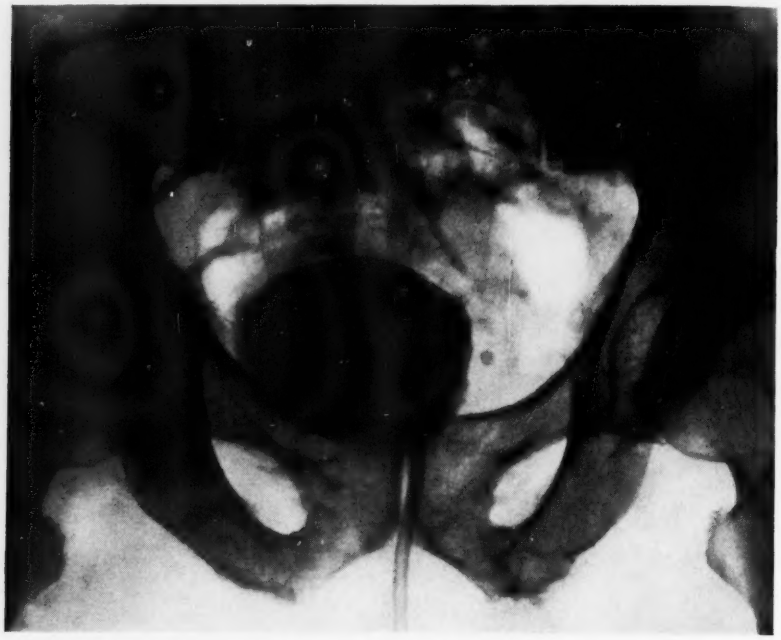


FIG. 4.—Cystogram. Bladder on right side of pelvis, but no prostatic intrusion effects seen.

pubic wound healed, but a few months later, cystoscopy revealed viable tumor tissue at the site of the original vesical tumor. The tumor was thoroughly fulgurated and six additional radon seeds were introduced transcystoscopically. Two months later, the patient again had urinary frequency, urgency and hematuria, with infected urine, and five ounces of residual urine. Recurrent tumor was seen, and also median prostatic lobe hypertrophy. The tumor was fulgurated and transurethral resection of the prostate was performed, January 7, 1942. The clinical course was progressively downhill. He died in March, 1942, five years after the original abdomino-perineal resection, and ten months after discovery of the bladder tumor. There was no necropsy.

Comment: The findings at exploratory celiotomy might indicate that the bladder lesion was a solitary local recurrence or metastasis. This, of course, would be most unusual. The surgeon had proposed total cystectomy, with ureteral implantation into the skin, but the patient had refused.

Case 4.—Hospital No. 461544: L. S., was a 33-year-old man, with symptoms of rectal bleeding, weight loss, and sense of mass in rectum, for at least four months.

A typical abdomino-perineal resection was performed, August 16, 1939, for a low rectal tumor. The rectum was very adherent to prostate, seminal vesicles and membranous urethra. The pathologic report was "infiltrating adenocarcinoma of rectum. Adjacent lymphatics show carcinomatous involvement." One year later, he was readmitted to the hospital for frequency and difficulty in urination, and attacks of acute retention. Cystometric studies showed a slightly hypotonic curve. The residual urine was 18 ounces. On intravenous pyleography, a normal right upper urinary tract, with incomplete filling on the left side, was found. Cystoscopy showed lateral lobe intrusion and some bar formation. Following cystoscopy he had an attack of acute pyelnoephritis and a *B. proteus* bacteremia. On September 26, 1940, a transurethral resection was performed. The pathologic report was "fragments of prostatic tissue and bladder showing infiltrating adenocarcinoma identical with that of previously resected adenocarcinoma of the rectum." The patient's urinary symptoms improved, and he was discharged to the Dept. of Radiotherapy, but received only four treatments. He died at a municipal institution four months later (one and one-half years after the rectal resection). The cause of death was given as "metastases" to bladder and prostate, chronic pyelonephritis, and uremia.

Comment: This is another typical case illustrating bladder and prostatic involvement after rectal resection, with death from upper urinary tract infection, *etc.*, probably due to ureteral obstruction by recurrent neoplasm in the pelvis.

Case 5.—Hospital No. 477854: H. W., was a 53-year-old male, with a six-month history, who presented himself to the Mt. Sinai Hospital, September 11, 1940, with a tumor of the rectum near the prostate. The biopsy showed "infiltrating colloid adenocarcinoma." An abdomino-perineal resection was performed. The convalescence was uneventful except for a temporary urinary leak from an operative perforation of the urethra. The extirpated specimen showed lymph node metastases. Seven months later, the patient was cystoscoped because of frequency, dysuria and pyuria. The cystoscopist thought that only an acute cystitis was present, but he biopsied a small posterior vesical wall protrusion. This showed "fragments of bladder mucosa and mucus cell carcinoma." No treatment was given. The patient entered the Montefiore Hospital, August 26, 1941, and died, February 8, 1942 (17 months after the original operation). The necropsy diagnoses were: Recurrent colloid carcinoma, with invasion of bladder, prostate, seminal vesicles, and innominate bone, and pressure on great vessels of pelvis. Metastases to the mesenteric lymph nodes, testis, epididymis and left adrenal. Compression of the right ureter by tumor tissue, with pyoureter, pyelonephritis, and perinephritis. Congenital absence of left kidney. Acute splenic tumor. Some parenchymatous degeneration of viscera. Congestion and edema of lungs.

Comment: From the postmortem findings, it does not appear likely that any of our known therapeutic aids could have helped this patient. However, at the time of the late postoperative cystoscopy, the perineal wound was still open. I think radium might have been applied to the bladder and pelvis through this wound, in an attempt to check the local recurrence although I do not know of any case helped in this way.

Case 6.—Hospital No. 407608: M. C., a 55-year-old woman, was operated upon December 30, 1935. A one-stage abdomino-perineal resection was performed. The tumor was three inches above the anus and infiltrated the rectal wall. The report was adenocarcinoma with lymph node metastasis. The patient developed severe perineal and back pain and, after 16 months, showed an extensive invasion of the whole pelvis

and bladder. Thoracic chordotomy was performed, resulting in some relief of the pain. The patient died 19 months after the original operation.

Case 7.—Hospital No. 433675. D. E., was a 41-year-old man with symptoms of four months duration. He had a large ulcerating mass in the ampulla of the rectum. On December 6, 1934, a parasacral resection of the rectum, with preservation of the sphincter and restoration in continuity, was performed. The pathologic report was adenocarcinoma with one lymph node involved. The entire specimen measured 6 x 8 cm., and was occupied by an annular carcinoma with normal rectum 6 mm. on either side. The tumor, grossly, infiltrated all coats and perirectal fat. The patient was well for three and one-third years, when pelvic pain and constipation developed. At the site of anastomosis, a recurrent cancer was found and confirmed by biopsy. On April 18, 1938, an abdomino-parineal resection with permanent sigmoid colostomy was performed. The liver was free of metastases. The rectum was adherent to the concavity of the sacrum, and the recurrent rectal cancer involved the prostrate, necessitating removal of the posterior aspect of the prostate and capsule. The histologic report showed adenocarcinoma in the rectal submucosa and prostatic tissue, with one involved lymph node. Patient's recovery was satisfactory, but eight months later patient entered hospital with symptoms of recurrent profuse urinary bleeding. The intravenous pyelogram showed bilateral upper urinary tract dilatation. Cystoscopy showed cancerous invasion of the base of the bladder. He died five and one-half years after the first operation, from repeated attacks of severe vesical hematuria. There was no postmortem.

Comment: Although the same postoperative sequelae often follow initial radical resection, partial resection with preservation of the anal sphincter is considered an inadequate procedure for this disease. Certainly, the patient stands a better chance against local residual cancer with later active recurrent growth in the pelvis if the initial operation is wide and complete.

Case 8.—Hospital No. 487704: N. S. was a 48-year-old male, with symptoms of rectal bleeding for several months due to a cancer palpable 5 cm. above the anal margin. On September 14, 1936, a first-stage Lahey operation was performed. No metastases were present. On October 3, 1936, the second-stage combined operation was undertaken. The tumor measured 6 x 4 x 2 cm. The pathologic report was "adenocarcinoma." The specimen was prepared by the oil of wintergreen method, but none of nine lymph nodes which were found showed metastastic involvement. The postoperative convalescence was uneventful. Approximately 15 months later (January 24, 1938) the patient was readmitted because of right loin pain, fever, and hematuria. The urologic study indicated a normal left upper urinary tract, and a functioning infected hydroureteronephrosis on the right side. A right ureteronephrectomy was performed. The kidney, histologically, showed evidence of acute and chronic pyelonephritis and ureteritis. The patient was readmitted to the hospital, May 9, 1941, about five years after the original resection because of difficulty in voiding, burning, nocturia and hematuria. Cystoscopy showed a large solid neoplasm on the floor of the bladder. The neoplasm was biopsied and fulgurated. Histologically, the biopsy showed fragments of bladder wall infiltrated by adenocarcinoma. The patient was discharged two weeks after admission. On October 22, 1941, a large papillary neoplasm was seen cystoscopically on the right side of the floor of the bladder overlying the right ureter orifice. This was again fulgurated. Later, because of severe right loin pain, a chordotomy was performed at the Neurological Institute. On the fourth admission (January 4 to 12, 1942) vesical neck obstructive symptoms were present, with a residual urine of 16 ounces. On cystoscopy, three calculi were seen on the floor of the bladder. At the site of the previously treated tumor, there was some irregular elevation of the

mucosa. The stones were crushed, and three grams of tissue were resected from the inferior sphincter. No evidence of malignancy was found. He left the hospital voiding without difficulty. The fifth admission was terminated by the patient's death, May 18, 1942, about six years after the two-stage resection. He had evidences then of diffuse metastases, particularly in the liver. The picture was one of gradual deterioration with increasing bladder irritability. There was no postmortem examination.

Comment: Although not proved, local residual recurrent cancer around the right ureter at the bladder may have led to the early infected right hydroureteronephrosis.. The tumor was slow growing, and vesical involvement did not become pronounced until three and one-half years later. Even if radical extirpation of the bladder had been performed, this probably would not have influenced the development of hepatic metastases.

Case 9.—Hospital No. 483687: J. S., a 40-year-old male, was admitted to the Mt. Sinai Hospital, April 6, 1941, and a one-stage abdomino-perineal resection was performed on the day of admission. The tumor was situated about 2.5 cm. from the anus; perirectal tissues were indurated, and the left seminal vesicle was adherent to the rectum. The pathologic report was "infiltrating adenocarcinoma of the rectum, with involvement of many lymph nodes." About six months after discharge, dysuria, pyuria and complete urinary retention was found to be due to irregular intrusion of tissue at the vesical neck. A transurethral resection was carried out, December 27, 1941. The histologic report was "fragments of prostatic tissue infiltrated by adenocarcinoma." Comparison of the slides of the rectal carcinoma and the resected prostatic tissue led Doctor Klemperer to the conclusion that the prostatic cancerous infiltration was secondary to the rectal cancer. Because of persistent urinary symptoms, including some incontinence, the patient entered the Urologic Service of one of the city hospitals. Here, another transurethral prostatic resection was performed, and the report of the removed tissue was adenocarcinoma of the prostate. A bilateral orchidectomy was performed, February 13, 1942, and a permanent suprapubic cystotomy established, March 14, 1942, for persistent urinary difficulty. He was admitted to the Montefiore Hospital, April 12, 1942. The serum "acid" phosphatase was 3.6 King-Armstrong units. Although no metastases were found on admission, he later developed cord compression from upper cervical vertebral metastases and died, October 6, 1942, one and one-half years after the rectal extirpation. A limited necropsy was done. The anatomic diagnoses were as follows: Adenocarcinoma infiltrating prostate, with direct extension to bladder wall, pelvic soft tissues and left ischium. Right pyonephrosis and perinephric abscess. Right and left pyoureter. Right and left pyelonephritis. Subacute cystitis. Acute splenic tumor.

Comment: This case offered considerable difficulty in determining the primary site of the cancer, inasmuch, as the autopsy findings could be interpreted either as a primary cancer of the rectum or prostate. No prostatic tissue could be recognized either grossly or microscopically in the dense scar-like tumor masses which occupied the pelvis. In view of the definite surgical diagnosis of cancer of the rectum, and particularly the age of the patient (41 years), cancer of the rectum seemed the more probable. A review and comparison of all the surgical and autopsy material from the three hospitals was made, which led to the conclusion that the slides from the resected rectum, prostate, and autopsy were histologically similar and, in view of all the evidence, a final diagnosis of primary cancer of rectum

was made. The importance of differential diagnosis is based on the possible therapeutic efficacy of orchidectomy in cancer of the prostate, as originally advocated by Huggins.[3] For future cases, two possible aids are suggested to distinguish between primary cancer of the prostate and secondary cancer of the prostate from a primary rectal cancer. The first is the serum acid phosphatase determination.[4] Primary prostatic cancer with metastases usually gives an elevated serum acid phosphatase, while in primary rectal cancer with or without prostatic involvement, the determination should be normal.[5] A second histologic aid may be in the use of a new staining method of transurethrally resected prostatic tissue. This method, described by Gomori,[6] demonstrates acid phosphatase activity in tissue sections. Prostatic cancer which contains large amounts of acid phosphatase stains strongly positive, as compared with a rectal cancer infiltrating the prostate gland which should, on the information at hand, not stain heavily. However, further studies will have to be made on these suggested laboratory aids.

While on the subject of prostatic cancer, attention should be called to the fact that the differential diagnosis between it and a low, anterior wall, primary rectal cancer may be difficult. Kickham and Bruce[2] mention four cases of prostatic cancer simulating rectal cancer in which the presenting symptoms were rectal. We have had several similar experiences.

Concerning the frequency of this sequela of lower urinary tract malignant infiltration following abdomino-perineal resection, I cannot give exact figures from our material. There have been no recently published statistics from this institution relating to the follow-up observations of the operative cases of carcinoma of the rectum. One of the reasons is that the grouping of these cases has changed hands several times, so that not enough time has elapsed for a proper evaluation. A recent study[7] of the immediate results of the surgical treatment of cancer of the rectum on the wards of the hospital for a four-year period showed that 85 patients were subjected to operation. Fifty-one were operable. In 36 cases, a one-stage abdomino-perineal resection was carried out, with six deaths, or an operative mortality of 16.6 per cent. In the remaining 15 operable cases, various other procedures were undertaken. The three ward cases, which have been detailed above, are from the group of 30 patients who survived the one-stage abdomino-perineal resection. Therefore, in this group the incidence of late involvement of bladder, prostate or seminal vesicles would be at least 10 per cent. However, our figures are too small to be reliable. Ewert[8] analyzed the urologic complications in 452 cases of extirpation of the rectum. The rectal lesion initially involved the ureter in 11 cases, the bladder in 23 cases, and the prostate in 19 cases. He states that in five patients in whom rectal resection was done, the growth invaded the prostate after operation, producing obstruction which required bladder neck resection. However, the number of cases followed postoperatively, or the length of follow-up is not mentioned.

It is outside of the scope of this communication to discuss in detail the

urinary complications of surgically treated or untreated cancer of the rectum or sigmoid. The literature on this subject is ample. From a urologic viewpoint, however, it is worth remembering, as pointed out by Schwartz and Bergman,[9] that urinary symptoms believed to be due to a lesion in the urinary tract are not infrequently secondary to a primary lesion in adjacent bowel, either neoplastic or inflammatory.

SUMMARY

Nine instances of late invasion of the bladder and/or prostate and seminal vesicles following abdomino-perineal resection for cancer of the rectum or recto-sigmoid are reported. The management of these cases is described. One of the patients appears to have been remarkably helped by radiotherapy and transurethral resection. The sequelae under discussion may occur more often than formerly realized. Upper urinary tract complications are found very frequently in the later stages of cancer of the lower bowel. Although cancerous invasion of the lower urinary tract after abdomino-perineal resection appears to render the situation hopeless palliative urologic or surgical measures and radiation therapy should often be attempted, with the expectation of prolonging life.

REFERENCES

1 Jones, T. E.: Complications of One-Stage Abdomino-Perineal Resection of Rectum. J. A. M. A., **120,** 104–106, September, 1942.

2 Kickham, C. J. E., and Bruce, N. H.: Urological Complications in Malignant Disease of the Rectum. Journal of Urology, **41,** 541–556, April, 1939.

3 Huggins, C., Stevens, R. E., Jr., and Hodges, C. V.: Studies on Prostatic Carcinoma; II. The Effects of Castration on Advanced Carcinoma of the Prostate Gland. Archives of Surgery, **43,** 209–233, August, 1941.

4 Sullivan, T. J., Gutman, B., and Gutman, A. B.: Theory and Application of the Serum "Acid" Phosphatase Determination in Metastasizing Prostatic Carcinoma: Early Effects of Castration. Journal of Urology, **48,** 426–458, October, 1942.

5 Gutman, A. B.: Personal communication.

6 Gomori, G.: Distribution of Acid Phosphatase in the Tissues under Normal and Pathologic Conditions. Archives Pathology, **32,** 189–199, August, 1941.

7 Garlock, J. H., Ginzburg, L. and Glass, A.: Complications and Causes of Mortality of the Surgical Treatment of Carcinoma of the Colon and Rectum. To be published.

8 Ewert, E. E.: A Comparative Analysis of the Urological Complications following Large Bowel Surgery. Journal of Urology, **46,** 764–776, October, 1941.

9 Schwartz, J., and Bergman, H.: Urologic Aspects of Carcinoma of the Colon and Rectum. Urological and Cutaneous Review, **41,** 3–7, 1937.

COCHLIOMYIA AMERICANA INFESTATION IN MAN

CASE REPORT

R. O. Pearman, M.D., and L. Haseman, Ph.D.*

St. Joseph, Mo.

Cochliomyia americana C. and P. is the name given to the New World screw-worm fly by Cushing and Patton[1] to separate this dangerous form, which feeds on the tissues of living animals, from the blowfly (*Cochliomyia macellaria*) which is more particularly a scavenger fly.

The screw-worm fly is native to the warmer sections of the Americas, but on its own power or by the movement of infested animals may spread into the "corn belt," to cause serious damage to livestock and other warm-blooded animals, including man. The adult fly may readily be mistaken for a medium-sized blowfly. It has a deep greenish-blue metallic color with yellow, orange or reddish face, and three dark stripes on the dorsal surface of the thorax. Unlike the blowfly, however, it is normally attracted to fresh cuts or bloody wounds rather than to purulent sores. Its larvae bore into and destroy the healthy tissue surrounding the wound, which may prepare the wound for a subsequent attack by the common blowflies. On livestock, wire cuts, dehorning wounds, castration cuts, navel wounds, and other skin injuries are the usual sources of screw-worm entrance. On man, entrance occurs most commonly through the ears, nostrils or exposed surface wounds, usually while the individual is asleep or unconscious.

In the last few years, the screw-worm fly has been of increasing importance as a pest of livestock in the Middle West. While, presumably, it is unable to withstand the winters, new infestations have been established each summer by the shipment of infested animals from the south and by the northward migration of the adult flies. In 1941, the pest spread from centers of introduction so as to cover entire counties and involve hundreds of animals. With frosts coming so late, as in the last two open falls, the pest has been able to continue to breed later than usual, so as to attack animals and man throughout October. In southern California,[2] this menace is present the year round, being more prevalent in the late fall and winter.

Along with this increase of its importance to livestock has developed its real menace to man. Any untreated cut, scratch, blister, insect bite, or other raw wound on the surface or any irritation in the nostrils, ears and eyes of man or beast may serve as an attraction to the fly. The female fly may deposit a few hundred eggs at the rate of 60 per minute, and with the eggs hatching in about a day, an untreated wound may soon become heavily infested with maggots. The young worms promptly begin to penetrate and destroy the tissues, eating in by means of the powerful oral hoods, and causing

* Professor of Entomology, University of Missouri, Columbia, Mo.

a burning, pinching type of pain, characteristic urine-colored discharge, and a most disagreeable odor. The maggots may become full-fed in four to a maximum of ten days. The adult worms drop out of the wound, bury in the ground, and after a week in the pupa stage emerge as a fly which soon mates and more eggs are deposited. This means that several broods of the pest may develop during the summer months.

Wallace[3] states that there appears to be a slight rotary motion as the worms busy themselves with their destructive feeding, and that their motility is due to the circular spines around their bodies, which are similar to the thread on screws, and from which they get their characteristic name.

Literature[3, 4, 5, 6] dealing with the screw-worm fly refers to various cases of infestation in man. Most of these cases were undoubtedly screw-worm infestations, although the maggots were rarely saved and identified as those of the screw-worm fly by experts in this field.

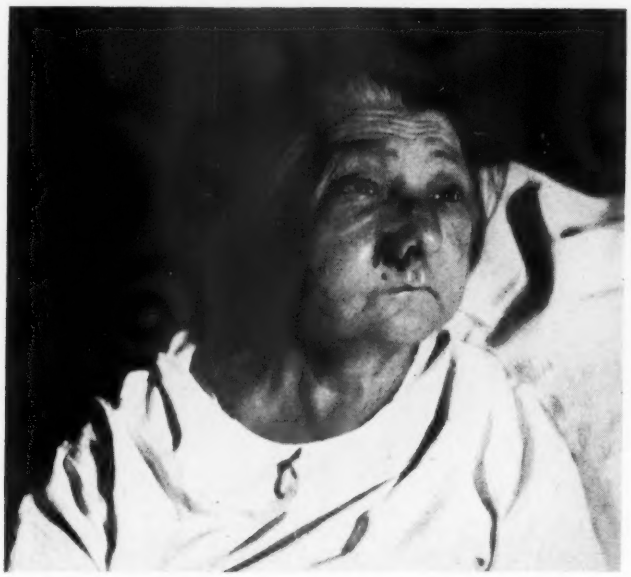


FIG. 1.—Patient after the removal of the larvae. Note the gangrenous area lateral to the right nostril.

August 12, 1941, one of us (R. O. P.) removed 80 apparently full-fed maggots from a lesion of the cheek of an elderly woman, which were identified by C. T. Greene, D. G. Hall, and later by E. F. Knipling,[7] Federal authority on screw-worm infestation, as definitely the larvae of the screw-worm fly, *Cochliomyia americana* C. and P.

Case Report.—D. M., age 58, married, was admitted to St. Joseph Hospital, August 11, 1941, with what, at first, appeared to be an abscess of the right side of her face. She said that she had been bitten on the right cheek, five days before admission to the hospital, by a spider or fly. The following day the right side of her face itched and became swollen and painful. The swelling became larger and the pain increased in intensity; and the night before admission to the hospital she coughed up a larva, and developed a bloody discharge from the nose and mouth. Ten hours

before admission, she noticed a gangrenous area about the size of a dime at the site of what she thought to be the original bite.

Physical examination was essentially negative except for a marked swelling of the right side of her face surmounted by a gangrenous area 1.5 cm. in diameter, and located 1.75 cm. lateral to the right nostril. Temperature 101° F., pulse 90, respirations 20. W.B.C. 11,350, neutrophile 80%, lymphocytes 20%. R.B.C. 3,250,000, hemoglobin (Sahli) 61%, blood Wassermann positive. Roentgenologic examination of sinuses: Both maxillary and right anterior ethmoid sinuses were moderately cloudy—not dense, but sufficient to indicate pathologic residue. Chest: Negative.

The patient was anesthetized with intravenous pentothal sodium, the nose and right cheek were painted with tincture of methiolate. The lesion was opened through the gangrenous area by means of a hemostat. A few larvae were obtained, accompanied

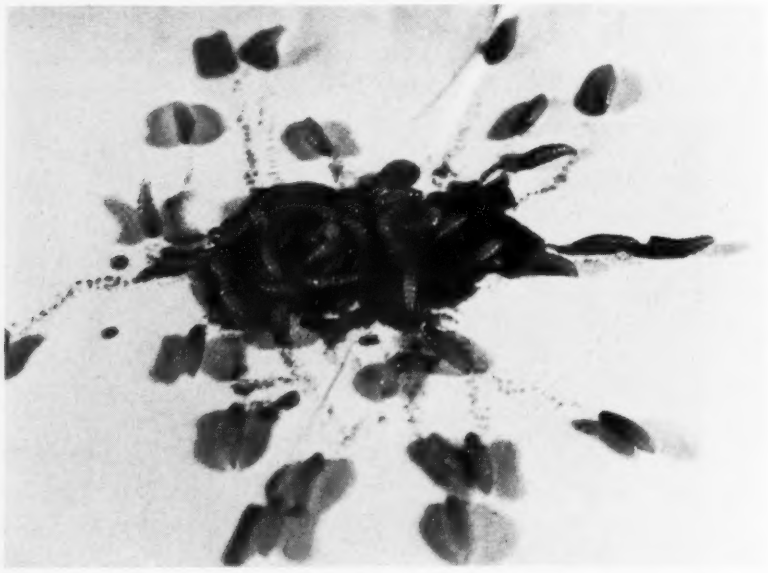


FIG. 2.—A few of the larvae obtained (X½). They were so active that several crawled out of the photographic field before the picture could be taken.

by approximately an ounce of foul smelling serosanguineous discharge. Upon further exploration of the lesion, many more larvae were observed spreading out in a fan-shaped manner and burrowing into the ragged, bloody and somewhat honeycombed tissue at the periphery of the lesion. These larvae were removed individually with a thumb forcep (Fig. 1). A total of 80 larvae were removed from the lesion. The ragged lining of the resulting cavity was curetted away down to healthy appearing tissue. The cavity was noted to open into the right nasal passage, and the nasal septum was perforated. The maxilla was roughened, but the process had not broken into the right maxillary sinus. The curettings and several pieces of tissue from the edge of the opening were saved for pathologic diagnosis. The cavity was swabbed out with tincture of methiolate and packed with vaselined gauze. *Pathologic Report:* Inflammatory tissue.

Most of the larvae were preserved in alcohol, although a few were kept in hopes that adult flies might emerge, but they failed to do so. The larvae were very active, of the usual maggot shape, 12 to 15 mm. in length and 2 to 2.5 mm. in diameter (Fig. 2), and gray-white in color with a tint of reddish-brown due to the tissue exudate which filled their intestinal tracts. The larva had 12 body segments, each of which was encircled by a narrow ring of minute spines, giving the larva the resemblance to

a screw. The head of the larva was rather pointed and was provided with an oral hood surmounted by two hook-like projections. The larger end of the larva was provided with two brownish plates through which it is supposed to breathe.

A volatile poison such as chloroform or benzol will kill the larvae and aid in their removal, especially in inaccessible lesions. However, in this case, the use of such an agent was not necessary.

The patient was discharged, August 19, 1941, markedly improved. Two months later, the area was completely healed and the patient appeared normal. No antiluetic therapy had been given in the interim, because the patient refused to come back to the clinic for treatment.

The patient lives in a poor and very unsanitary part of the city, about a mile from the stockyards and packing houses. The house opens directly onto the sidewalk and is a dark shack crowded between two similar houses. The windows were not screened, but the front entrance had an old, dilapidated screen door. The backyard was low, littered with trash and overgrown with weeds. At the time she was supposed to have been bitten by an insect, the hide of a butchered goat hung in the coal house, and the place was alive with flies. She also reported flies in the house. She stated that she did not take naps in the daytime, and that she did not have any sores or scratches on her face nor any nasal discharge at the time of the supposed bite.

Those of us who saw the case were of the opinion that the fly must have deposited its eggs in her right nasal cavity, and that the maggots worked out from there to form the lesion in the cheek. This would seem to be the logical explanation; for if there were no breaks in the skin on the cheek, the fly must have laid its eggs in the patient's right nostril. The fly probably gained access to the house, and in the early morning while the patient slept, deposited the eggs in her nostril, where perhaps unknown to her she may have had some irritation. Then the supposed spider or insect bite on the cheek was probably merely pain due to the first activity of the young maggots, some five days before they were removed at operation.

COMMENTS

(1) With the spread of the screw-worm infestation among cattle, more cases in humans may be expected in the near future.

(2) Maggots from any wound should always be saved for identification by an entomologist.

(3) Wounds should always be kept covered to protect them from flies.

(4) Persons afflicted with sores about, or discharges from, the aural or nasal cavities should sleep only in well-screened houses, and should keep the areas protected from possible attack by flies when outside.

(5) A volatile poison (chloroform or benzol) will kill the larvae and aid in their removal, especially from inaccessible lesions.

REFERENCES

[1] Cushing, E. C., and Patton, W. S.: Studies on the Higher Diptera of Medical and Veterinary Importance. Cochliomyia Americana, the Screw-worm Fly of the New World. Ann. Trop. Med. and Parasitol., **27**, No. 4, 539–551, November, 1933.

[2] McCapes, A. M.: California Polytechnic College, San Luis Obispo, Calif. Personal communication.

[3] Wallace, W. R.: Screw-worm Infestation: (Case Report). J. South Carolina M. A., **32**, 213–215, September, 1936.

[4] Craig, C. F., and Faust, E. C.: Clinical Parasitology, Philadelphia, Lea and Febiger, 1940, p. 613.

[5] Turnbull, F. M., and Franklin, L. B.: Aural Infestation with Screw-worms. J. A. M. A., **120**, 117–119, September 12, 1942.

[6] Knipling, E. F., and Rainwater, J. T.: Species and Incidence of Dipterous Larvae Concerned in Wound Myiasis. J. Parasitol., **23**, 451–455, October, 1937.

[7] Knipling, E. F.: Dept. of Entomology, Bureau of Agriculture, Washington, D. C. Personal communication.

BRIEF COMMUNICATIONS

UMBILICAL DISCHARGE IN ACUTE APPENDICITIS

REPORT OF TWO CASES

PHILIP D. ALLEN, M.D., AND ROBERT A. JOHNSON, M.D.

NEW YORK, N. Y.

FROM THE CHILDREN'S SURGICAL SERVICE, BELLEVUE HOSPITAL, NEW YORK, N. Y.

INVOLVEMENT of the umbilicus secondary to inflammatory disease of the appendix is uncommon. Cullen,[1] in his treatise on the diseases of the umbilicus, states that he has never seen this structure involved in appendicitis.

Umbilical discharge may occur as the result of spontaneous rupture of an appendiceal abscess resulting in an entero-umbilical fistula or it may result from a generalized peritonitis secondary to inflammation of the appendix. A case will be presented illustrating each of these two conditions.

Purulent drainage from the umbilicus secondary to a generalized peritonitis has been reported by many writers, including Heurtaux,[2] Vaussy,[3] and Gauderon.[4] The peritonitis in most instances has been classified as idiopathic. The pneumococcus was the most frequent organism found, and the condition occurred most often in young females. Haggard[5] reported umbilical discharge occurring in a case of peritonitis in which the clinical signs indicated acute appendicitis as the basic pathologic process, but this was not proved by operation or autopsy.

Case 1.—Hosp. No. 194701: R. S., white, female, age two and one-half years, was admitted to the hospital, August 14, 1941, with a history that five days before she had had an upper respiratory infection, anorexia, vomiting and abdominal pain. Three days after onset the child had two generalized convulsions and was referred on the fifth day to Willard Parker, a hospital for contagious diseases, with a tentative diagnosis of meningitis. She was at once transferred to Bellevue Hospital, with a diagnosis of perforated appendix and spreading peritonitis.

On admission, the child was in a prostrate condition, with a temperature of 103° F., pulse 124, respirations 40. Positive physical findings were limited to the abdomen, which was markedly distended, with moderate tenderness throughout. No masses were palpable. Rectal examination revealed induration but no definite mass.

The white blood count was 9,500, with 75 per cent polymorphonuclear leukocytes, 24 per cent lymphocytes, and 1 per cent monocytes. There was 1+ albumen in the urine. The hematocrit was 44, and the plasma protein 5.5 grams.

Supportive therapy consisting of small blood transfusions, oxygen and sulfonamides were given. Clinically, the child improved, and on the fifth hospital day the temperature was 100° F., pulse 110, white blood count 14,150. Two days later thick, creamy pus began to drain from the umbilicus in small amounts. Under local anesthesia, a McBurney incision was made and a large amount of thick, purulent material was evacuated from the peritoneal cavity. Drains were inserted without further exploration and the wound was packed open with vaselined gauze. A right lower lobe pneumonia

developed postoperatively, and purulent material continued to drain from the umbilicus and the operative wound. The patient expired on the seventh postoperative day.

Necropsy.—Limited to the abdomen. The peritoneal cavity was the site of a purulent plastic peritonitis. There were numerous loculated abscesses one of which, in the region of the umbilicus, contained several hundred cubic centimeters of yellowish-green pus. Examination of the bowel revealed that the cecum opened into the free peritoneal cavity through the eroded opening of the appendiceal stump. The remainder of the appendix was not found.

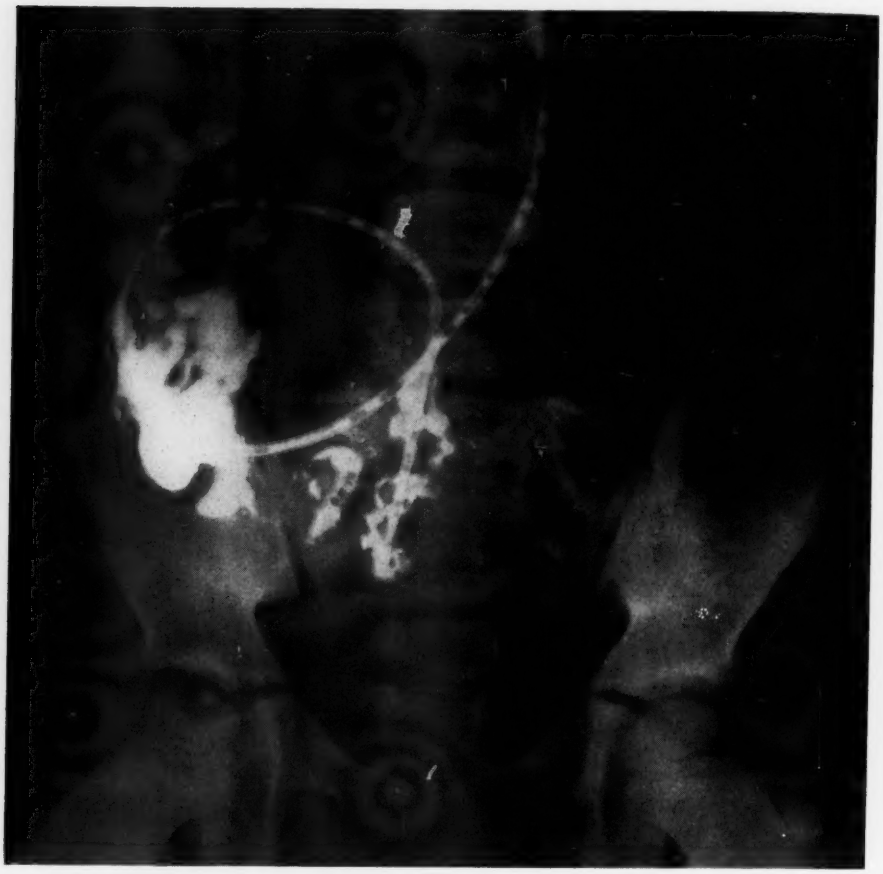


FIG. 1.—Roentgenogram showing the lipiodal visualization of the umbilical sinus.

The drainage of an appendiceal abscess through the umbilicus with the establishment of an entero-umbilical fistula has been reported in the literature by Kelly and Hurdon,[6] Vaussy,[3] Bryant and Hine.[7] These case reports present clinical histories which would seem to implicate the appendix, but operative or autopsy evidence is lacking.

Case 2*.—Hosp. No. 37410-42: L. B., white, female, age four, was admitted to Bellevue Hospital, July 9, 1942, with the chief complaint of discharge from the umbilicus, of one week's duration. The mother stated that 15 days prior to admission the child had had a transitory attack of abdominal pain, without vomiting or distur-

* Presented before the New York Surgical Society, November 11, 1942.

bance of bowel habit. This brief episode did not force the child to go to bed, and she continued to play about in the usual manner.

Physical examination was negative except for the umbilicus, in the center of which was a small pouting area presenting an opening about five millimeters in diameter. The surrounding skin was eroded and the discharge was yellowish-green in color, without fecal odor. The abdomen was soft, without tenderness, spasm or palpable mass. Rectal examination revealed a small mass at the tip of the examining finger.

The temperature was 99° F., on admission. The white blood count was 7,200, with 70 per cent polymorphonuclear leukocytes, and 30 per cent lymphocytes. Gross and microscopic examination of the urine was negative. Examination of the discharge for free hydrochloric acid was negative. Twenty cubic centimeters of methylene blue was injected into the tract, but none appeared in the urine. Ten cubic centimeters of lipiodol was then injected into the tract and roentgenograms visualized the terminal ileum, cecum and ascending colon (Fig. 1). A diagnosis of Meckel's diverticulum, with a patent fistulous opening through the umbilicus was made, and operation was performed on the fifth hospital day.

Operation.—Through a right rectus muscle-splitting incision, a firm mass, covered with omentum, was found to be attached to the anterior abdominal wall about five centimeters below the level of the umbilicus. On dissecting free the omentum there was found to be encapsulated in it a sloughing appendix. The cecum was identified and the base of the appendix transected, the stump inverted, and what remained of the appendix was removed in a retrograde fashion. The fistulous tract leading to the umbilicus was laid open, curetted, and packed with vaselined gauze.

The postoperative course was uneventful. The opened sinus tract healed in slowly from below, and was entirely healed when the child was discharged on the twentieth postoperative day.

The pathologic laboratory confirmed the diagnosis of acute and chronic inflammation of the appendix.

SUMMARY

These two case reports have been presented, with the operative and autopsy findings, to illustrate the uncommon occurrence of umbilical discharge secondary to inflammatory disease of the appendix. This may occur either as the result of generalized peritonitis of appendiceal origin, as illustrated by Case 1, or by the spontaneous drainage of an appendiceal abscess, with the formation of an entero-umbilical fistula, as in Case 2.

REFERENCES

[1] Cullen, T. S.: The Umbilicus and Its Diseases. W. B. Saunders Co., Philadelphia, p. 296, 1916.

[2] Heurtaux, A.: Phlegmon sous ombilical. Bull. et Mein de la Soc. de Chir. de Paris n. s., **3**, 641, 1877.

[3] Vaussy, F.: Des phlegmons sous peritoneaux de la paroi abdominale anterieure. These de Paris, No. 445, 1875.

[4] Gauderon, A. D.: De la peritonite idiopathique aigue des enfants de sa terminaison par suppuration et par evacuation du pus a travers l'umbilic. These de Paris, No. 148, 1876.

[5] Haggard, W. D.: Quoted by Cullen, T. S.[1]; The Umbilicus and Its Diseases. W. B. Saunders Co., Philadelphia, p. 305, 1916.

[6] Kelly and Hurdon: The Vermiform Appendix and Its Diseases. 1905, p. 202. W. B. Saunders Co., Philadelphia.

[7] Bryant and Hine: Medical Times and Gaz., **2**, 460, 1878.

SPONTANEOUS RUPTURE OF A NORMAL SPLEEN

CASE REPORT

CAPTAIN ANGUS D. MCLACHLIN, R.C.A.M.C.

TORONTO, CANADA

FROM THE DEPARTMENT OF SURGERY, UNIVERSITY OF TORONTO, AND SURGICAL SERVICE, TORONTO GENERAL HOSPITAL, TORONTO, CANADA

THE NORMAL SPLEEN is not uncommonly ruptured by trauma. On occasion, the loss of blood at time of injury may be slight because the laceration is speedily occluded by a thrombus. In some instances this thrombus may proceed to healing by fibrosis. In other cases it may be detached by further injury or may soften and be freed spontaneously. A second and much more severe intraperitoneal hemorrhage may then take place. Jackson[1] recorded a case of this type in which the severe hemorrhage occurred 28 days after the initial injury. The term "exploding spleen" has been coined to describe this second phenomenon. A review of the early literature on traumatic rupture of the normal spleen and of the history of splenectomy was made by Barnes.[2]

Preexisting disease of the spleen is a strong factor in determining rupture by trauma. A blow to an abdomen containing a large, friable malarial spleen might well result in a fatal hemorrhage from that organ, a fact taken advantage of by ingenious assassins of the middle ages.

Susman[3] found record of spontaneous rupture of the spleen in malaria, typhoid, pregnancy, parturition and the puerperium, leukemia, hemophilia, acute infections, tuberculosis, and even more rarely in typhus fever, relapsing fever, cystic degeneration, malignant growth, hydatid disease, infarction, torsion, abscess and varices.

Spontaneous rupture of an apparently normal spleen is much less frequent. Zuckerman and Jacobi[4] collected 20 genuine and seven doubtful instances in the period prior to 1937. Since then, Dudgeon[5] has contributed another case. Recent European journals have contained articles on the subject, but are not available on this continent. The purpose of this report is to present another example of spontaneous rupture of an apparently normal spleen.

Case Report.—B. K., male, age 46, lumberjack, suffered from generalized crampy abdominal pain with nausea and vomiting and some tenderness in the right lower abdomen for one week, a year prior to admission. He received no medical attention at that time, and on recovery felt entirely well until one week before coming into the hospital. During that week he developed symptoms suggestive of a subacute intestinal obstruction. Physical examination on admission was in accordance. There was no abdominal rigidity or tenderness, and no masses could be made out. On roentgenologic examination, the plain films revealed moderately distended loops of small bowel, and the barium enema showed a fixed and irregular terminal ileum, suggestive of an inflammatory disease. The distension cleared up rapidly after admission, and he re-

mained in bed, with no complaints, and with no elevation of temperature, increased pulse rate or unusual W.B.C. count.

Twelve days after admission, without premonitory symptoms, he developed sudden, severe generalized abdominal pain, followed in a few moments by pain behind the left shoulder tip. The pain was increased by any shifting of his body. On examination, he appeared acutely ill, with ashen color and clammy skin. The abdomen was slightly distended and uniformly tender. There was uniform guarding of the abdominal wall, but not true board-like rigidity. Free fluid was easily demonstrated, but Ballances' test was not attempted. The abdomen was silent. Rectal examination revealed extreme tenderness high up, but was not otherwise helpful. There was no free air in the abdominal cavity demonstrable roentgenologically. The pulse was 130; B.P. 80/60; temperature 99.2° F. per rectum; and W.B.C. 23,000.

After administration of morphine, 250 cc. of plasma, and 600 cc. of whole blood, the general condition of the patient improved sufficiently to make celiotomy feasible. The abdominal cavity was entered through a short midparamedian incision on the right side. Bloody fluid and blood clots presented immediately. A rapid manual examination revealed that there were more blood clots in the left upper abdomen than elsewhere. The incision was extended upward, and the spleen delivered into the wound. The splenic capsule had been stripped up and split open, and there was blood clot adherent to the splenic pulp. The vessels were ligated separately at the hilum and the spleen removed. The liver was normal in size, color and consistency. There were adhesions between loops of small intestine in the lower abdomen, but further exploration did not seem warranted at this time. About 1500 cc. of blood clot and another 100 cc. of bloody fluid was removed from the peritoneal cavity; the wound was closed without drainage; and the patient returned to bed to an oxygen tent. Recovery was free from complications. The hemoglobin was 33 per cent the day following operation, with an admission level of 80 per cent. The platelet count was 473,000 at five days, and still 410,000 two weeks after operation. Blood smear, differential W.B.C., and van den Bergh level were normal, and the Wassermann was negative.

Because of the still undiagnosed lesion in the lower abdomen, the abdominal cavity was opened again five weeks after the splenectomy. The small bowel "tie-up" was found due to a former acute appendicitis. This was straightened out and the appendix removed. Convalescence was once again uneventful.

Pathologic Report: This specimen consists of a small, markedly contracted, relatively avascular spleen, which measures 8 x 6 x 3.5 cm. On the diaphragmatic surface of the spleen the capsule has been stripped up by an effusion of blood. Portions of blood clot are still adherent to the outer part of the splenic pulp. Sections of the spleen reveal it to be firm and contracted, and liver-like in consistency, almost no blood exuding from the cut surface. The malphighian corpuscles and trabeculae are difficult to make out. Serial gross sections of the spleen fail to reveal any infarct or other area of softening to account for the subcapsular hemorrhage. The vessels at the hilum of the spleen fail to reveal any abnormality. No traumatized area, and no zone definitely identifiable as the site from which the hemorrhage occurred is present.

Microscopic sections of this spleen fail to reveal any information as to the mechanism resulting in the extensive subcapsular hemorrhage. The spleen is markedly contracted, so that the sinusoids and the other vascular channels contain almost no blood. The arterioles are thickened throughout. The trabeculae appear normal. The capsule, where stripped up, shows a slight chronic inflammatory infiltration of lymphocytes.

Discussion.—The correct diagnosis was not reached before operation. A proven lesion at the terminal ileum made it seem the most likely source of the general peritoneal irritation. The development of pain in the left shoulder

tip soon after onset of the general abdominal pain suggested that the accident had occurred in the upper abdomen. The short midabdominal incision, capable of extension in either direction, was an admission of this uncertainty in diagnosis.

The clinical findings were not entirely in keeping with perforation of a hollow viscus. The fall in blood pressure and rise in pulse rate seemed out of proportion to the other clinical findings and might have suggested that blood loss played a part in the picture. Free fluid was demonstrated both by transmission of fluid wave and by shifting dullness. Ballance's sign was not tried, and might have solved the problem.

The cause of a spontaneous hemorrhage from an apparently normal spleen in difficult to explain. In the absence of a rent in the splenic pulp it seems likely that the initial hemorrhage was between the splenic pulp and the capsule. A logical sequence of events would then be a gradual elevation of the splenic capsule, rupture of the capsule and then massive intraperitoneal bleeding following on the relief of pressure. No blood clot of any age was found, and a slight chronic inflammatory infiltration of lymphocytes in the stripped up splenic capsule is the only evidence that the whole process did not occur in rapid order.

The etiology of the initial hemorrhage is even more perplexing. A minor blow to the abdomen just sufficient to rupture a small vessel deep to the capsule and initiate the process is an attractive theory. In opposition to this, the patient had been confined strictly to bed for 12 days prior to the accident, and had been very quiet during the preceeding week. On careful questioning he could recall no blow to the abdominal wall during the previous several months that had been severe enough to be remembered.

Susman[3] noted that vague gastro-intestinal symptoms were sometimes present in the period before rupture of a normal spleen. He believed that congestion of the portal vein and its radicals might force blood between the splenic pulp and capsule and finally force the latter to give way. The period of subacute intestinal obstruction immediately before rupture in this case could well have produced a portal congestion. In contradiction of this theory, spontaneous rupture of the spleen has not been reported in portal cirrhosis or thrombosis, where portal congestion should be much more severe.

Wohl[6] observed that the malphighian bodies and capsule of the spleen showed thickening and hyaline changes at a comparatively early age. He considered that spontaneous rupture of the apparently normal spleen was due to a local degeneration in the wall of a blood vessel. In the case presented here the arterioles were thickened, but these changes seemed within the normal range.

Perisplenic adhesions which fixed the spleen in position might, conceivably, predispose to injury, but were not present here.

In conclusion, no definite cause can be assigned to this spontaneous rupture of an apparently normal spleen. Its occurrence soon after an episode

of subacute intestinal obstruction is some evidence that venous congestion might play a rôle in the etiology.

SUMMARY

1. A case of spontaneous rupture of an apparently normal spleen is presented.

2. There was no tear in the splenic pulp, and a subcapsular hematoma, with stripping of the splenic capsule and final rupture into the peritoneal cavity, seemed the likely sequence of events.

3. No adequate cause for the hemorrhage was found, but the accident occurred during convalescence from a subacute intestinal obstruction, and is some evidence that venous congestion may play a rôle in the initial hemorrhage.

I wish to thank Dr. Roscoe R. Graham for permission to publish this case and Professor W. L. Robinson for the pathologic report.

REFERENCES

1 Jackson, T. S.: Rupture of the Spleen: Report of an Unusual Case. Surg. Gynec. & Obst., **41,** 331–332, September, 1925.

2 Barnes, A. F.: Subcutaneous Traumatic Rupture of the Normal Spleen. ANNALS OF SURGERY, **59,** 597–609, 1914.

3 Susman, M. P.: Spontaneous Rupture of Spleen. Br. Jour. Surg., **15,** 47–54, July, 1927.

4 Zuckerman, I. C., and Jacobi, M.: Spontaneous Rupture of Normal Spleen. Arch. Surg., **34,** 917–928, May, 1937.

5 Dudgeon, H.: Spontaneous Rupture of Spleen. South. Med. Jour., **34,** 1247–1249, December, 1941.

6 Wohl, M. C.: Spontaneous Rupture of Spleen. ANNALS OF SURGERY, **82,** 246–249, August, 1925.

BOOKS RECEIVED

First Aid Surgical and Medical. By Warren H. Cole, M.D., and Charles B. Puestow, M.D., M.S., Ph.D. New York: D. Appleton-Century Company, Inc., 1942.

Synopsis of Traumatic Injuries of the Face and Jaws. By Douglas B. Parker, M.D., D.D.S. St. Louis: C. V. Mosby Co., 1942.

Traumatic Surgery of the Jaws. By Kurt H. Thoma, M.D. St. Louis: C. V. Mosby Co., 1942.

The Time of My Life. By Harry Carlos De Vighne, M.D. Philadelphia and New York: J. B. Lippincott Co., 1942.

Lymph Node Metastases. By Grantley Walder Taylor, A.B., M.D., F.A.C.S., and Ira Theodore Nathanson, M.S., M.D. New York: Oxford University Press, Inc., 1942.

Transactions of the Southern Surgical Association, Fifty-fourth Annual Meeting. Edited by Alton Ochsner, M.D. Philadelphia and New York: J. B. Lippincott Co., 1941.

Religion and Health. By Seward Hiltner. New York: Macmillan Co., 1943.

Manual of Oxygen Therapy Techniques. By Albert H. Andrews, Jr., M.D. Chicago: The Year Book Publishers, Inc., 1943.

Atlas of Ovarian Tumors. By Gemma Barzilai, M.D. New York: Grune and Stratton, 1943.

Transactions of the Sixty-third Meeting of the American Surgical Association. Edited by Walter Estell Lee, M.D. Philadelphia and New York: J. B. Lippincott Co., 1942.

Treatment of Fractures. By Guy A. Caldwell, M.D., F.A.C.S. New York and London, Eng.: Paul B. Hoeber, Inc., Medical Book Department of Harper Brothers.

EDITORIAL ADDRESS

Original typed manuscripts and illustrations submitted to this Journal should be forwarded prepaid, at the author's risk, to the Chairman of the Editorial Board of the ANNALS OF SURGERY

Walter Estell Lee, M.D.
1833 Pine Street, Philadelphia, Pa.

Contributions in a foreign language when accepted will be translated and published in English.

Exchanges and Books for Review should be sent to James T. Pilcher, M.D., Managing Editor, 121 Gates Avenue, Brooklyn, N. Y.

Subscriptions, advertising and all business communications should be addressed

ANNALS OF SURGERY
East Washington Square, Philadelphia, Pa.